ILLNESS SELF-MANAGEMENT AMONG ADULTS LIVING WITH ANCA SMALL
VESSEL VASCULITIS

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ABSTRACT
CAROLYN T. THORPE: Illness Self-Management among Adults
Living with ANCA Small Vessel Vasculitis
(Under the direction of Robert F. DeVellis)

This dissertation examines illness self-management among adults living with ANCA-associated small vessel vasculitis (ANCA-SVV). Manuscript #1 describes the development and evaluation of the Vasculitis Self-Management Scale (VSMS), a self-report measure of self-management among adults with ANCA-SVV. Manuscript #2 characterizes patients' attitudes and beliefs, including perceived barriers and facilitators, about ANCA-SVV self-management, and examines their relationship to self-management. Manuscript #3 explores spousal accommodative behavior as a supportive process that facilitates self-management. Specifically, it tests whether patients' perceptions of general and/or illness-specific spouse support mediate the relationship between spousal accommodative behavior and patients' self-management.

The VSMS was administered via mail questionnaire, along with measures of socio-demographics, clinical factors, psychosocial factors, and self-management attitudes and beliefs. The sample for manuscripts #1 and #2 consisted of 205 patients, while the sample for manuscripts #3 included 159 married patients.

The final VSMS assessed eight domains: medication adherence, health services adherence, infection avoidance adherence, diet adherence, exercise adherence, symptom monitoring adherence, appropriate adjusting of activities in response to fatigue or symptoms, and prompt reporting of new symptoms or side effects to a health professional. Analyses demonstrated good evidence of internal consistency and test-retest reliability, and mixed evidence of construct validity.

With few exceptions, patients' perceived difficulty and number of perceived barriers were negatively associated with self-management, while greater perceived importance predicted higher levels of self-management. Some specific barriers, but not facilitators, were negatively associated with self-management.

Spousal constructive accommodation was positively associated with general, but not illness-specific, support. General support was positively associated with medication adherence, health services adherence, and appropriate adjusting of activities in response to fatigue and symptoms. Spouse retaliation was negatively associated with illness-specific, but not general, support. Illness-specific support was positively associated with medication adherence, infection avoidance adherence, and appropriate adjusting of activities in response to fatigue and symptoms. Formal tests of the indirect effects of spousal accommodative behavior on self-management via spouse support approached but did not reach statistical significance.

Findings suggest that the VSMS is a promising method for assessing illness self-management in adults living with ANCA-SVV. Potential avenues for future intervention efforts with this population are considered.

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CHAPTER ONE: INTRODUCTION

Individuals who are diagnosed with ANCA-associated small vessel vasculitis (ANCA-SVV) face a daunting array of psychosocial challenges. In addition to living with an unpredictable and potentially fatal prognosis, patients and their families are asked to undertake complicated treatment regimens that come with considerable risk of serious side effects (Jayne 2003) and require substantial role adjustment and lifestyle changes (Koutantji, Pearce et al. 2000). Yet leading researchers have noted the paucity of research on the psychosocial aspects of living with vasculitis. In an editorial outlining some of the unanswered questions about the psychological aspects of vasculitis, Koutantji, Pearce, and Harrold (2000) suggested that a major goal of future research should be to evaluate coping, functioning, and quality of life issues in patients living with vasculitis, all issues that have been under-studied in this patient population.

One of these issues not previously studied is patients' illness self-management, or the daily tasks an individual performs, with the collaboration and guidance of health care providers, to control or reduce the impact of disease on his/her physical health status. Patients with ANCA-SVV are often asked to perform a variety of health behaviors, including taking medication, using health services, following diet and exercise guidelines, monitoring symptoms, taking actions to avoid infection, and responding appropriately to illness symptoms. The lack of research is troublesome, given that non-adherence to recommended treatment behaviors is common across different chronic illnesses (DiMatteo 2004), and could

lead to ineffective treatment, serious complications, and even death in patients with ANCA-SVV. Complicating the study of illness self-management in these patients is the lack of an existing self-report measure that can reliably, validly, and conveniently measure adherence to the treatment behaviors commonly recommended by physicians to ANCA-SVV patients, and those that are generally recommended for all ANCA-SVV patients. Development of such a measure would allow for the empirical study of important influences on illness self-management in this population, which would in turn provide guidance for interventions that are tailored specifically for vasculitis patients to improve their self-management behavior. Thus, the first purpose of the dissertation was to develop and test a self-report measure of ANCA-SVV self-management with a sample of adults living with ANCA-SVV, and gather evidence of its reliability and validity. A second and related purpose was to characterize patients' attitudes and beliefs, including perceived barriers and facilitators, about performing self-management behaviors among adults living with ANCA-associated small vessel vasculitis (ANCA-SVV), and determine if these attitudes and beliefs were associated with levels of self-management behavior.

In addition to building on the current state of the literature on quality of life issues among this specific patient population, the proposed research aims to expand our knowledge regarding the impact of marital relationships on self-management behavior in chronic illness more generally. Previous research with other chronically ill populations suggests that social relationships may act as one important influence on illness self-management. Researchers have repeatedly found that patients' perceptions of social support provided by close others in their lives often acts as a facilitator of self-management behavior in patients living with a variety of chronic illnesses (Gallant 2003; WHO 2003; DiMatteo 2004). In this body of

research, social support has typically been defined as a resource that individuals perceive to be available or provided in close relationships (Cohen, Gottlieb et al. 2000).

Unfortunately, these consistent observations about the beneficial effect of naturally-occurring social support on self-management behavior have not translated into clear knowledge about how to intervene to improve support in close relationships. Spousal support interventions specifically targeted at improving patients' self-management behavior are few in number. Related interventions designed to mobilize the support of key network members (often spouses) in the hopes of changing health behaviors and improving psychological adjustment to chronic illness have yielded modest and mixed results (Lichtenstein, Glasgow et al. 1986; Black, Gleser et al. 1990; Cutrona and Cole 2000; Riemsma, Taal et al. 2003; Park, Schultz et al. 2004). One reason often cited for mixed intervention results is the simplistic treatment of social support as a perceived resource and a resulting lack of understanding about the mechanisms underlying its provision and receipt (Lassner, 1991).

In response to these findings, a new approach to studying social support and health has begun to emerge from the field of relationship science. This approach conceptualizes support as a dynamic *process* resulting from the motivations and skills of the provider and receiver of support as well as specific interactions in close relationships, rather than simply a *resource* (DeVellis, Lewis, et al., 2003). Such an approach could help inform future spousal support interventions by offering more specific guidance on how to favorably change patients' perceptions of spousal support, which has proven to be very difficult in many couples-focused interventions (Lichtenstein, Glasgow, et al., 1986). Two constructs from this

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approach – transformation of motivation and accommodative behavior – are a promising

alternative to understanding how support may affect patients' illness self-management.

   The construct of transformation of motivation comes from Interdependence Theory

(Rusbult and VanLange, 1996), which is commonly used to guide research on outcomes in

close relationships (e.g., relationship maintenance or dissolution) (DeVellis, Lewis, et al.,

2003). Interdependence Theory suggests that spouses experience individual outcomes as

well as shared outcomes related to the goals of their relationship, and that spouses mutually

influence each others' outcomes. A central concept in Interdependence Theory is

transformation of motivation (Yovetich and Rusbult, 1994), or when an "individual

internalizes social interactions from another person, and thus acts in pro-relationship, health-

enhancing ways rather than acting in a purely self-interested manner" (DeVellis, Lewis, et al.,

2003). When transformation of motivation has occurred, each partner's goals become the

other partner's goals, and each partner's behavior reflects these mutual goals. One

commonly used indicator of whether transformation of motivation has occurred is spouses'

accommodative behavior, which refers to the extent partners react constructively rather than

destructively when the other acts in a relationship-threatening way (i.e., with hostility or

criticism) (Rusbult, Verette, et al., 1991). Given that patients often cite negative interactions

with spouses as a barrier to patients' carrying out illness management behaviors (Maclean,

1991; Clark, Janz, et al., 1994; Handron and Leggett-Frazier, 1994; Clark and Nothwehr,

1997), increased accommodative behavior on the part of spouses may act as a facilitator to

self-management behavior among chronically ill individuals. Therefore, this research was

undertaken with a third primary purpose: to determine if spouses' accommodative behavior

functions as a supportive process that facilitates ANCA-SVV self-management behavior.

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This dissertation is organized around three separate manuscripts that correspond to the three primary research purposes described above. Chapter 2 includes a review of the literature and a description of how this literature informed the specific aims and conceptual models underlying the three manuscripts in the dissertation. Chapter 3 explicitly states the specific aims and hypotheses of each manuscript. Chapter 4 (manuscript #1) reports on the development and initial testing of the new measure of ANCA-SVV self-management, including evaluation of its dimensionality, reliability, and validity. Chapter 5 (manuscript #2) characterizes the patients' attitudes and beliefs, including perceived barriers and facilitators, about performing the self-management behaviors relevant to ANCA-SVV, and explores whether these attitudes and beliefs are associated with self-reported levels of self-management behavior. Chapter 6 (manuscript #3) investigates the relationships among spousal accommodative behavior, perceived spousal support, and patients' self-management behavior, and evaluates perceived spousal support as a mediating process by which accommodative behavior may influence self-management behavior. The final chapter then summarizes the main findings of the dissertation and discusses their public health implications, along with the overall strengths and limitations of the research. Directions for future research and intervention are also described.

CHAPTER TWO: BACKGROUND AND SIGNIFICANCE

Overview of Chapter

In this chapter, I begin by describing ANCA-associated small vessel vasculitis (ANCA-SVV), its epidemiologic burden, and its treatment. I then describe the concept of illness self-management, and the closely related construct of treatment adherence, and why it is a concern in ANCA-SVV. This section is followed by a discussion of issues related to the measurement of self-management behavior and a rationale for my approach to measuring ANCA-SVV self-management in this research. The section on self-management concludes with a discussion of its correlates that are common across chronic illnesses, which provides a rationale for the selection of construct validity variables assessed in this study. The third major section of this chapter discusses social support, and specifically perceptions of spouse support, as a determinant of self-management behavior in chronic illness. The traditional theoretical approach, which treats support as a resource to be provided by spouses and received by patients, is described along with evidence of its relevance to self-management behavior from observational and intervention studies. I then offer a critique of this traditional approach and argue that viewing support as a relationship process is a more promising and useful approach in terms of intervention development. Finally, I introduce the concepts of transformation of motivation and accommodative behavior as potential relationship processes that underlie patients' perceptions of spousal support, and present a conceptual model of how spousal accommodative behavior may influence patients' illness self-management behavior via perceived spousal support.

ANCA-associated Small Vessel Vasculitis

Primary systemic vasculitis is a group of relatively rare, potentially fatal, autoimmune

conditions, characterized by inflammation and necrosis of blood vessels (Watts and Scott,

1997). Wegener's granulomatosis (WG), microscopic polyangiitis (MPA), and Churg-

Strauss syndrome (CSS) are three types of small vessel vasculitis that are often grouped

together in the scientific literature as ANCA-associated small vessel vasculitis (ANCA-SVV)

because they affect the body in similar ways. WG, MPA, and CSS are associated with the

presence of anti-neutrophil cytoplasmic antibodies (ANCA). All three conditions cause

inflammation and necrosis of small blood vessels and medium-sized arteries primarily in the

respiratory tract and kidneys (Watts and Scott, 1997) and are treated using similar

immunosuppressive medication regimens (Watts and Scott, 1997; Jayne, 2003).

The overall annual incidence of primary systemic vasculitis and its specific sub-types

varies by geographic region and study methods used. In the United Kingdom, the annual

incidence of ANCA-SVV was estimated to be 39 per million (Watts, Carruthers, et al., 1995);

in a German study, the annual incidence ranged from 9.5 to 16 per million over a five-year

period (Reinhold-Keller, Herlyn, et al., 2005). In both the British and German studies, WG

was found to be more common than both CSS and MPA; however, MPA was found to be

more common than WG in Spain (Reinhold-Keller, Herlyn, et al., 2005). ANCA-SVV

affects children and adults of all ages, and men and women alike, yet incidence increases

with age and peaks in the elderly (Watts, Lane, et al., 2000). WG, MPA, and CSS appear to

be more prevalent in Caucasians than in racial/ethnic minorities (Mahr, Guillevin, et al.,

2004).

Characteristics of ANCA-associated Small Vessel Vasculitis

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While the nature and severity of morbidity varies both within and across the three types of ANCA-SVV, all three are potentially fatal, relapsing diseases characterized by an often progressive yet unpredictable course (Koutantji, Pearce, et al., 2000). All three conditions can cause permanent organ damage, most often to the respiratory tract and kidney, as well as the nerves, heart, and skin. Until recently, rapid death from ANCA-SVV was likely; one study from the 1980s found that 82% of WG patients died within a year of diagnosis (Fauci, Haynes, et al., 1983). The introduction of effective immunosuppressive medications into vasculitis treatment regimens in the 1990s transformed vasculitis from a rapidly fatal disease into a chronic illness with which patients often live for many years (Koutantji, Pearce, et al., 2000). Although there is no cure for ANCA-SVV, treatment with immunosuppressive medications typically induces remission in 80-100% of patients (Langford, 2003). Patients may experience full remission for many years, partial remission with "grumbling" disease activity (sometimes referred to as "low disease state"), and/or minor or major relapses (Jayne, 2003). Despite the effectiveness of immunosuppressive medications in inducing remission, the risk of relapse and morbidity from both the disease and its ongoing highly toxic treatment remains high (Hoffman, Drucker, et al., 1998). Rates of relapse range across studies but have been reported to be as high as 50% by five-years of follow-up (Bacon, 2005) and is more common in WG than in MPA (Bacon, 2005). In one recent United States study, 57% of WG patients experienced a relapse and 22% experienced a relapse that was classified by the researchers as severe (WGET, 2005).

Treatment Regimens for ANCA-SVV

Treatment regimens for ANCA-SVV are long-term, complex, frequently altered by physicians to address changes in disease activity, and associated with serious side effects.

Together with their physicians, patients typically agree to follow a demanding regimen which includes a variety of self-care tasks, including medication-taking, obtaining vaccinations, regular use of health services and communication with their physicians, and a range of other lifestyle changes. These regimens are often adjusted based on current disease activity and the occurrence of side effects and complications. Patients may also receive varying amounts of assistance from family and friends in performing these self-care tasks.

The primary strategy for treating ANCA-SVV consists of long-term therapy with several different types of immunosuppressive medication. Initial remission is often induced by treatment over several months, or even years, with cytotoxic medications (e.g., cyclophosphamide and methotrexate) and corticosteroids (Jayne, 2003). Renal disease may develop and require treatment with additional immunosuppressive agents (Jayne, 2003) and less often, dialysis and kidney transplant (Hoffman, Drucker, et al., 1998). Once full or partial remission is achieved, immunosuppressive medication regimens are sometimes changed to include less toxic doses or drug types, or even discontinued after several years of symptom-free remission (Jayne, 2003). However, the majority (e.g., 78%, according to Hoffman, et al., 1998) of ANCA-SVV patients remain on immunosuppressive medication such as prednisone, methotrexate, cyclophosphamide, and/or azathiprine for many years. Even those who discontinue immunosuppressive therapy may remain on medications to treat or prevent complications of ANCA-SVV and its treatment.

Preventing and treating immunosuppressive medication side effects and ANCA-SVV complications is indeed a very important component of ANCA-SVV self-care regimens. Side effects of immunosuppressive medications are numerous and can cause serious and irreversible problems (WGAI, 2003). Particularly serious complications of long-term

immunosuppressive therapy for ANCA-SVV include cystitis, bladder cancer, neuropathy, steroid-induced osteoporosis and cardiovascular disease, and serious infections (WGAI, 2003). One potentially life-threatening infection that is of special concern to ANCA-SVV patients on long-term immunosuppressive medication is *Pneumocystis carinii* pneumonia (Jayne, 2003).

In the hopes of warding off these dangerous complications, ANCA-SVV patients are often prescribed additional medications, encouraged to obtain vaccinations for infectious diseases, and asked to keep regular appointments and communication with their physicians to closely monitor any developing complications. Commonly prescribed medications include long-term antibiotic therapy, medications to prevent bone loss, and folic acid (WGAI, 2003). Patients are also often encouraged to obtain regular vaccinations for influenza and pneumonia to reduce the risk of developing serious infections. In addition, patients (even those in remission) are also asked to monitor themselves continuously for symptoms and side effects, and attend regular medical visits as recommended by their physician (WGAI, 2003), so that any return of disease activity or development of side effects can be treated promptly and additional morbidity may be avoided.

In addition to taking medications and utilizing health services, ANCA-SVV patients may also be asked to make changes in various aspects of their lifestyle. For example, together with their physician, patients may decide to follow various recommendations for sleep, diet, and exercise, or may be encouraged to quit smoking and limit alcohol intake (WGAI, 2003). In addition, individuals experiencing ANCA-SVV-related complications may perform various behaviors to care for these complications (e.g., nasal irrigation, self-massage for relief of pain associated with peripheral neuropathy).

The above description reveals that self-care regimens for ANCA-SVV are quite complex and vary according to each individual's situation. However, little is known about the frequency with which patients are asked to carry out these behaviors, as well as the extent to which patients actually adhere to recommended self-management behaviors. This research, primarily that presented in Chapter 4, aimed to shed light on these issues.

<div align="center">Illness Self-management and Treatment Adherence</div>

The current accepted definition of illness self-management is the "day-to-day tasks an individual must undertake to control or reduce the impact of disease on physical health status" (Clark, et al., 1991). Patients assume these tasks with the collaboration and guidance of their health care providers (Clark, et al., 1991). A construct that is closely related to illness self-management is treatment adherence, or "the extent to which a person's behavior – taking medication, following a diet, and/or executing lifestyle changes – corresponds with agreed recommendations from a health care provider" (WHO, 2003). While many self-management tasks include adhering to physicians' recommendations, self-management is a broader construct of which treatment adherence is one aspect (Gallant, 2003). Self-management involves mastery of three types of activities: performing activities to manage the condition, making informed decisions about care, and applying skills to maintain adequate psychosocial functioning (Clark, et al., 1991). Specific self-management behaviors required by patients vary across conditions (Clark, et al., 1991). While many of these behaviors may be specifically recommended to patients by physicians based on patients' specific illness situations, self-management may also include behaviors not specifically recommended by physicians but those that are generally recommended for all patients with

the condition (e.g., attending to and promptly reporting new symptoms to a health professional, cutting back on daily activities when symptoms and fatigue flare up).

Evidence suggests that illness self-management behavior is a state rather than a trait. For example, there are no stable personality traits or attributes that reliably predict adherence to recommended health behaviors (Rand, 2000), and adherence typically varies over the course of an illness. Also, previous research has shown that rates of non-adherence vary across different self-care behaviors. For example, research with adult diabetes patients has repeatedly shown that an individual's adherence to one component of the diabetes regimen is not highly correlated with his/her adherence to other components of the regimen (Johnson, 1992; Toobert and Glasgow, 1994). In addition, patients' scores on measures designed to assess patients' general tendencies to adhere to medical recommendations have been shown to have relatively small correlations with their scores on measures assessing adherence to illness-specific behavioral recommendations (Hays, 1993). Thus, a measure of self-management that assesses performance of multiple behaviors, such as the one developed in this study, would likely be multi-dimensional in nature.

Performance of Self-management Behaviors in ANCA-SVV

Very little is known about the extent to which individuals living with ANCA-SVV perform recommended self-management behaviors, and next to nothing is known about the specific barriers to doing so that these patients face. However, what is known about adherence to long-term therapies in the context of other chronic illnesses suggests that adherence to behaviors commonly recommended for adults living with ANCA-SVV may be low. Investigations of treatment adherence in other chronic diseases repeatedly find that non-adherence is a problem common to most chronic diseases. A recent review by the World

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Health Organization (WHO, 2003) reported that only 50% of patients typically adhere to physicians' recommendations. A recent cross-disease meta-analysis found the average non-adherence rate across illnesses and type of behavioral regimen to be 24.8% (DiMatteo, 2004). Previous research has demonstrated that treatment non-adherence is more common when regimens are long-term, complex, frequently altered, and associated with severe side effects (WHO, 2003), which are all characteristics of ANCA-SVV treatment regimens. Furthermore, previous research on the impact of vasculitis on quality of life suggests that patients and their families face substantial challenges in adapting to and managing ANCA-SVV on a long-term basis (Koutantji, Pearce, et al. 2000).

Investigations of patients living with other chronic illnesses with similar psychosocial demands to those of ANCA-SVV (e.g., other forms of systemic vasculitis, certain types of cancer, and systemic lupus erythematosus) provide further evidence to suggest that non-adherence may be a problem in ANCA-SVV. For example, one French study of adherence to medication therapy in patients living with temporal arteritis (another form of systemic vasculitis) found that adherence to glucocorticoids, calcium, vitamin D, and bisphosphonates was 87%, 60%, 68%, and 51%, respectively (Le Gal, Queyrel, et al., 2003). Although relatively understudied, non-adherence to the same medications as those commonly used to treat ANCA-SVV (e.g., prednisone, cyclophosphamide, and other types of chemotherapy) has been documented as a problem in patients with various forms of cancer (Richardson, Marks, et al., 1988; Lebovits, Strain, et al., 1990; Partridge, Avorn, et al., 2002). One study found that 43% of women prescribed oral cyclophosphamide and/or prednisone for treatment of breast cancer reported either over-ingesting or under-ingesting the medications during a one-week period (Lebovits, Strain, et al., 1990). Another study found that only 30.8% of

African American patients with lupus and 23.4% of white patients with lupus reported taking their medication as prescribed all of the time, and around 10% of lupus patients reported that they *failed* to take their medication *all* of the time (Mosley-Williams, Lumley, et al., 2002). The same study also found that attendance at regularly scheduled clinic appointments for lupus was rather poor, with African American patients missing 27.4% of scheduled appointments and white patients missing 42.6% of scheduled appointments over a one-year period (Mosley-Williams, Lumley, et al., 2002). Given similarities in treatment regimens and the chronic, relapsing prognosis of these illnesses, these studies strongly suggest that non-adherence may be common in ANCA-SVV as well.

High rates of non-adherence among individuals living with ANCA-SVV could potentially be very problematic. While the consequences of medication non-adherence have not been formally studied in ANCA-SVV, adherence in chronic illness "…is a primary determinant of the effectiveness of treatment, because poor adherence attenuates optimal clinical benefit" (WHO, 2003, pp. 20). A recent meta-analysis of the relationship between treatment non-adherence and health outcomes revealed that adherence reduced the risk of poor treatment outcome by 26% across all illnesses studied, and that treatment adherence was more strongly associated with positive outcomes in chronic versus acute illnesses (DiMatteo, Giordani, et al., 2002). The results of one study of treatment and survival in WG patients with renal involvement demonstrate the potential for treatment non-adherence to result in poor clinical outcomes specifically in ANCA-SVV. In this study, non-adherence with immunosuppressive therapy was believed to be responsible for renal failure in at least two of the 25 patients followed for an average of 36 months (Andrassy, Erb, et al., 1991).

More research is clearly needed on the consequences of poor performance of the different self-care behaviors required by ANCA-SVV treatment regimens. However, we can reasonably assume that non-adherence in ANCA-SVV can potentially lead to significant morbidity and even mortality from both ANCA-SVV disease progression itself, as well as complications from its highly toxic treatment. For example, poor adherence to prophylactic antibiotic therapy could lead to the development of life-threatening infections.   Poor adherence to exercise and diet recommendations could lead to the development of osteoporosis and impaired quality of life. Furthermore, a patient's non-adherence may be mistaken as failure to respond to therapy and lead to unnecessary alterations in treatment regimens and wasteful diagnostic tests (Rand, 2000). Therefore, characterizing the extent to which ANCA-SVV patients adhere to the various behaviors involved in managing their condition and identifying patients' perceived barriers to performing these behaviors is important in order to determine if interventions to improve self-management of ANCA-SVV are needed.

Measurement of Illness Self-management

One obstacle to studying illness self-management in ANCA-SVV is the lack of an available instrument to reliably and validly measure it. The manuscript in Chapter 4 addresses this gap by reporting on the development of a new measure of ANCA-SVV self-management and evidence of the measure's reliability and validity.

While previous research on self-management behavior and treatment adherence has most often used thresholds for classifying patients into categories (e.g., adherent versus non-adherent, good versus bad self-management), these thresholds are often arbitrary and draw on little or no evidence to support them (WHO, 2003). Instead, a more useful conceptualization

of adherence (and thus, self-management) is that of a continuous phenomenon (WHO, 2003), where those who experience greater levels of adherence can be expected to have better health outcomes. This view of self-management was adopted for this study.

A patient self-report measure of self-management behavior was selected for development over other methods for multiple reasons. First, patient self-report is considered an essential component of any investigation into treatment adherence. While no "gold-standard" method currently exists for measuring adherence to treatment regimens in chronic illness and patient self-report has sometimes been shown to overstate one's actual performance of recommended behaviors (Partridge, Avorn, et al., 2002; WHO, 2003), the World Health Organization (2003, p. 5) has stated that "a multi-method approach that combines feasible self-reporting and reasonable objective measures is the current state-of-the-art measurement of adherence behavior." Second, the validity of self-report measures can be assessed, for example, in this study by examining how respondents' desire to present their behavior in socially desirable ways affects their scores on the self-management measure, or in future studies by comparing scores on the self-report measure to objective measures such as pill counts, electronic medication monitors, or chart reviews. Third, using a self-report measure allows for more convenient and less expensive assessment of adherence to a wide range of self-care behaviors relevant to the management of ANCA-SVV than would using a variety of objective measures to assess the performance of different self-care tasks (e.g., pill counts to assess medication-taking, clinic records to assess appointment-keeping, accelerometers to assess physical activity). Furthermore, because objective measures are not available or feasible for assessing some aspects of ANCA-SVV self-management (e.g., following dietary or infection avoidance recommendations, cutting back on daily activities in response to

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increases in illness symptoms), self-report is the only feasible way of assessing adherence to the full range of self-management behaviors required by most ANCA-SVV treatment regimens. Therefore, having an easy-to-use and inexpensive self-report measure of self-management behavior that covers the variety of self-care tasks central to ANCA-SVV treatment will be extremely useful for future investigations. Finally, assessing self-management behavior via self-report may also allow for a greater understanding of *why* patients sometimes fail to adhere to their regimens (Rand, 2000). For example, a self-report measure can easily be administered along with items that determine whether patients do not adhere unintentionally (e.g., because of forgetting or misunderstandings about regimens), or deliberately (e.g., because of a desire to avoid side effects or perceptions of treatment ineffectiveness). Assessing such perceived barriers to self-management behavior has the potential to guide future intervention efforts to improve self-management in this patient population.

While general, self-report measures of treatment adherence exist for use across patient populations (e.g., Hays's General Adherence Scale, 1993) regardless of the specific self-care tasks required by the chronic illness, it is important to develop an ANCA-SVV-specific measure of self-management behavior. Assessing adherence to specific self-care behaviors instead of global adherence captures variations in adherence across the different self-care behaviors required by ANCA-SVV treatment regimens, and is therefore much more informative for creating and evaluating future interventions to improve adherence. For example, if adherence is high for medication-taking but low for appointment-keeping and infection avoidance behaviors, future interventions can target more challenging behaviors and focus less on improving adherence to medication therapy that is already high.

Furthermore, the general self-report measures identified in the literature are limited to assessment of behaviors specifically recommended by physicians, to the exclusion of additional self-care behaviors generally considered appropriate for all patients with ANCA-SVV.

Correlates of Self-management Behavior

While there is no one theory as to the process by which illness self-management behavior is determined, the correlates and determinants of adherence to chronic disease treatment regimens have been studied extensively in the hopes of developing interventions to successfully intervene on these factors. Research on the factors associated with adherence and self-management behavior usually occurs in the context of a specific chronic illness; however, recent reviews of this literature (DiMatteo, Lepper et al. 2000; Gallant 2003; WHO 2003; DiMatteo 2004; DiMatteo 2004) reveal that several of these factors are consistently associated with self-management behavior across different chronic illnesses.

Several recent meta-analyses of the treatment adherence literature provide quantitative estimates of consistent, cross-disease associations between adherence to recommended behaviors and several demographic and psychosocial characteristics. Consistent with assertions that self-management behavior is not well-predicted by stable traits of individuals, DiMatteo (2004) found no relationship across studies of adult patients between adherence and patient age or gender, and only a weak association between patient educational achievement and adherence to chronic illness regimens (mean *r* effect size of 0.09; range of -.13 to .64). In a separate meta-analysis, DiMatteo (2004) found consistent, small-to-moderate positive relationships between perceptions of available social support from others and treatment adherence. The median *r* effect sizes for adherence and practical support,

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emotional support, and unidimensional social support were .27 (range -.22 to .75), .15 (range .00 to .37), and .20 (range -.06 to .60), respectively. In addition, DiMatteo, Lepper, and Croghan (2000) found an average moderate negative relationship (mean unweighted r equal to -0.27; range -.38 to -.17) for depressive symptoms and adherence.

Other systematic reviews of the treatment adherence literature, most notably the 2003 World Health Organization report on treatment adherence, suggest other factors that consistently predict adherence across disease types. In addition to depression and social support, the WHO report suggested that higher treatment complexity (e.g., more than one medication, or a more frequent dosing schedule), lower self-efficacy in performing regimen behaviors, longer disease duration, and having asymptomatic disease are negatively associated with treatment adherence. Quantitative estimates of the effect of these factors on treatment adherence across diseases were not available in this report and are therefore conservatively assumed to be small to moderate in magnitude.

In summary, what is known about the correlates of adherence to recommended behaviors in other chronic illnesses informed the study hypotheses related to the measure's construct validity, examined in Chapter 4; that is, the relationship of ANCA-SVV self-management behavior to demographic and psychosocial variables.

Social support as a Determinant of Illness Self-management Behavior

As noted above, social support has consistently been shown to predict self-management behavior across a variety of patient populations. Several theoretical perspectives have been applied to the study of social support as a determinant of treatment adherence. In this section, I will summarize and critique the dominant theoretical perspectives as well as provide a more detailed review of the empirical evidence supporting the relationship between social support

and treatment adherence. The section concludes with a presentation of an alternative approach to studying social support's influence on treatment adherence and ultimately, my conceptual model for my third manuscript, presented in Chapter 6.

Definitions of Social Support

Social support, defined broadly, refers to "any process through which social relationships might promote health and well-being" (Cohen, Gottlieb, et al., 2000). In studying the effects of social relationships on health, the first investigators drew upon Durkheim's observations that psychological distress and suicide were a direct result of breakdowns in individuals' social ties. This led them to focus on *structural* definitions of social support (e.g., marital status, social integration, or diversity of social contacts) when studying the ways in which social relationships might influence health. As the years passed, investigators began to explore how social relationships *function* to enhance or hurt one's health (Cohen, Gottlieb, et al., 2000). These functional investigations of the effects of social relationships on health led to two more narrow definitions of social support that have been adopted in a large proportion of the existing social support literature. It is these definitions that are most relevant to the current research.

In the first approach, social support is operationally defined as the social *resources* that patients perceive to be available to them (perceived support) or are actually provided to them (received support) by family members, friends, and acquaintances (Cohen, Gottlieb, et al., 2000). Support is often broken down further into several types, or dimensions: 1) emotional support, defined as the existence of someone who provides listening, caring, and acceptance; 2) tangible or instrumental support, defined as practical problem-solving help; 3) informational support, defined as information, guidance, and resources; 4) companionship,

defined as the availability of individuals with whom one can participate in social and leisure activities; and 5) validation or feedback support, defined as information about the normativeness of thoughts, feelings, and actions. These distinctions are thought to be important because certain types of perceived and received support may be more or less relevant to different types of stressors and behaviors or other outcomes (Wills and Shinar, 2000). Accordingly, social support has also been conceptualized both generally and with regard to specific stressors or disease conditions (Wills and Shinar, 2000).

In the second and less common approach, social support has been operationally defined as the interactional and relationship *processes* that facilitate coping with stressful life events (Reis and Collins, 2000). Stated a little differently, "support is the natural product of relationships exhibiting certain properties or involving certain types of interaction," and therefore can be linked to actual interpersonal events (Reis and Collins, 2000). The research on illness self-management behavior has mainly embraced the first definition, exploring much less often how relationship properties and specific interactions with close others affect self-management behavior among the chronically ill. A recent review of the social support and treatment adherence literature (DiMatteo, 2004) illustrates this bias: 68 studies defined support as a resource either perceived to be available or provided, while only 20 studies looked at relationship properties such as family cohesiveness or conflict.

Theoretical Perspectives

Regardless of how they define social support, most studies of the relationship between social support and treatment adherence has been relatively atheoretical, simply studying whether or not a relationship exists between perceived or received support and performance of recommended behaviors (Gallant, 2003), or between specific relationship properties such

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as family functioning or conflict and self-management behavior. Those researchers that have based their investigations in theory have primarily drawn on broad social support theories that are not specific with regard to the mechanisms by which social support influences self-management or health behavior more generally (Gallant, 2003). The three most commonly applied theoretical bases in studies of social support's influence on self-management behavior include the stress-buffering model of social support, the direct effects model, and an indirect model in which support influences behavior and health through psychological mediators (Gallant, 2003; DiMatteo, 2004). A fourth approach, that which defines support as the result of interactional and relationship processes, focuses more on the determinants of perceptions of support and has emerged in recent years as a promising theoretical perspective for studying the impact of social support on treatment adherence.

Stress-buffering Model

The stress-buffering model of social support has been the most influential and widely applied approach to studying the relationship between social support and health (Lakey and Cohen, 2000). The stress-buffering model proposes that the specific supportive actions by others, or *received* support, leads to improved coping, which in turn buffers acute and chronic stress and results in better health (see Figure 2.1). This model also proposes that the *perceived* availability of support leads to more positive appraisals of stress, which leads to better coping and ultimately, better health (see Figure 2.2). A more specific version of this model suggests that health behaviors, including treatment adherence and other chronic illness self-management behaviors, mediate the relationship between stress, coping, and health outcomes (Berkman and Glass, 2000). An important aspect of these models is that they

suggest that social support will have greater benefits for individuals experiencing high levels

of stress (Wills & Shinar, 2000).

Figure 2.1. Received support leads to improved coping, which moderates the relationship

between stress and health behaviors/outcomes. Adapted from Lakey & Cohen, 2000, p.31.

Received
Support

Coping

Stress   Self-management
behavior   Health
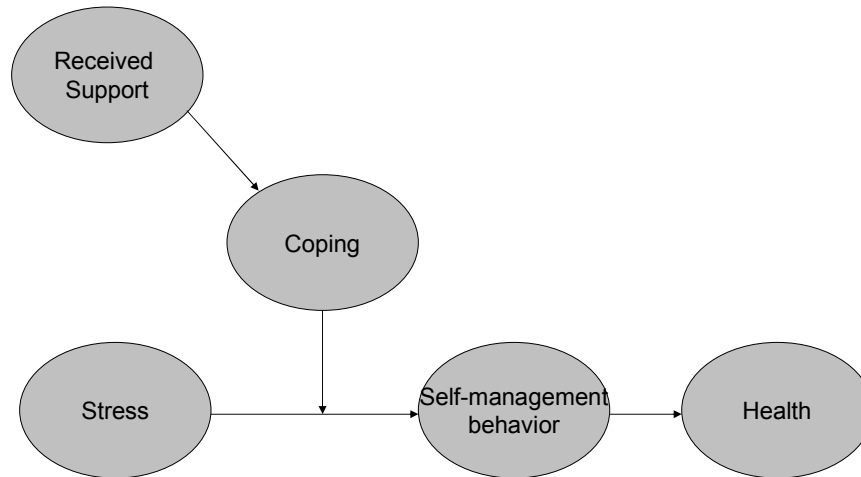
Figure 2.2. Perceived support leads to improved appraisals of stress, which moderates the

relationship between stress and health behaviors/outcomes. Adapted from Lakey & Cohen,

2000, p.32.

Perceived
Support

Appraisal

Stress   Self-management
behavior   Health
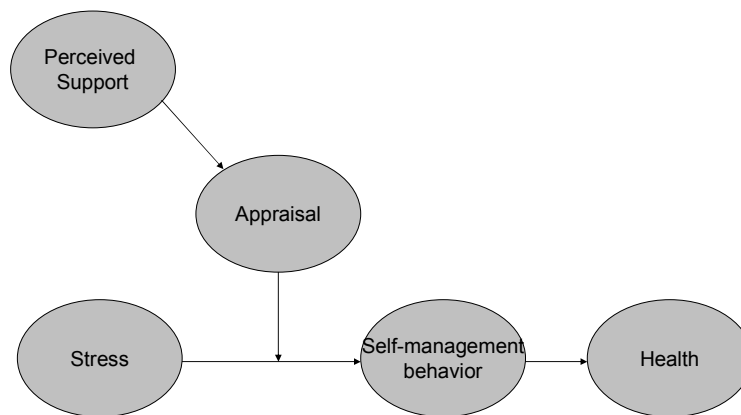
Direct Effects Model

In contrast, the direct effects model, also known as the main effects model, supposes that

social support influences behavior and health directly, regardless of one's level of stress

(Burman and Margolin, 1992). In terms of self-management behavior, this model (see Figure 2.3) would lead to the hypothesis that supportive others directly influence self-management by providing practical assistance with illness-management tasks (Gallant, 2003; DiMatteo, 2004).

Figure 2.3. Receipt of practical support directly influences self-management behavior, which in turn influences health outcomes.
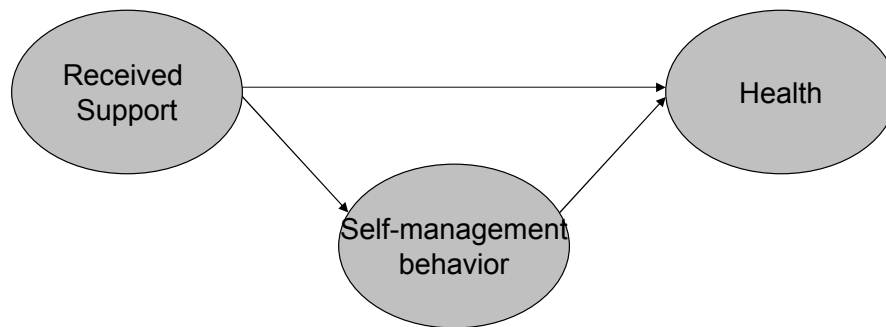
Indirect Effects Model

Other researchers have extended the direct effects model by hypothesizing that social support influences self-management behavior via psychological mediators that may include self-efficacy, psychological adjustment (e.g., depression), optimism, and/or self-esteem (Gallant, 2003; DiMatteo, 2004) (see Figure 2.4). Unfortunately, explicit tests of these mechanisms have been rare (Gallant, 2003; DiMatteo, 2004).

Figure 2.4. Perceptions of available support and/or receipt of support indirectly affect behavior and health via psychological mediators.

Psychological factors:
Self-efficacy
Psychological adjustment
Optimism
Self-esteem

Social Support

Health

Self-management behavior
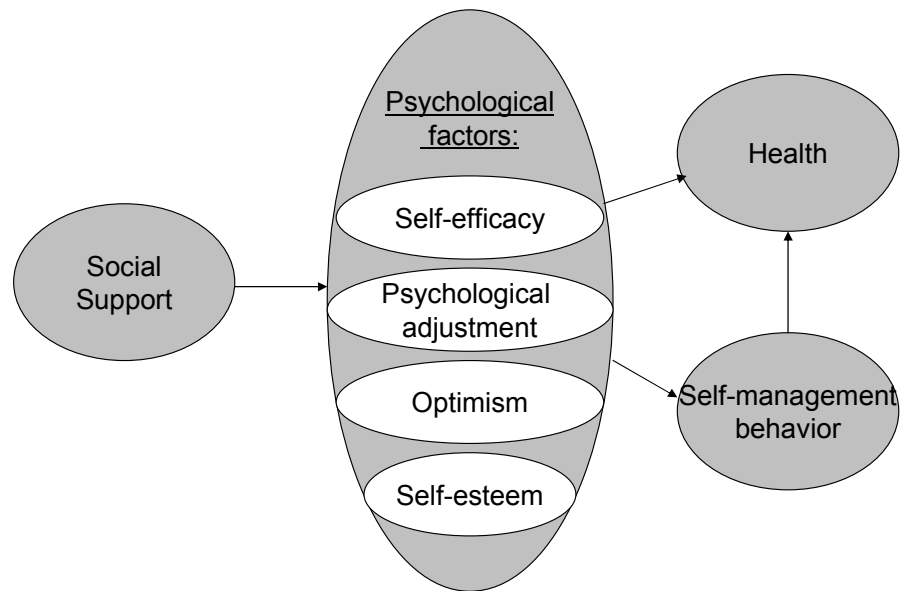
Relationship Processes Perspective

As described above, an alternative model (see Figure 2.5 below) of social support's influence on health has gathered momentum in recent years in the literature on health and behavior, and self-management behavior more specifically. This model is derived from the field of relationship science and focuses more on the determinants of individuals' perceptions of support from close others (Reis and Collins, 2000). The basic tenet of this model is that perceptions of social support are the direct result of relationships that are characterized by certain properties and patterns of interaction (Reis and Collins, 2000). That is, perceptions of one's social resources result from processes that occur in their interactions and relationships with close others. More specifically, this model proposes that features of individuals' social networks and the interpersonally-relevant predispositions of individuals within those

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networks drive the nature of interactions and features of relationships among network members. In turn, specific interactions and properties of relationships drive one's perception of support available to them, as well as one's interpretations of behaviors intended to be supportive. The implication of this model is that individual network members' predispositions toward interactions with others (e.g., empathy, attachment, social competence), properties of relationships (e.g., intimacy, love, conflict), and specific, observable interactions with network members all interact to influence individuals' subjective evaluation of support available and provided to them in close relationships. As in the other social support models, perceived support is then hypothesized to influence health behaviors such as illness self-management, and thus, health outcomes. The relationship processes approach focuses less on the mechanisms by which perceptions of support drive behavior and health and more on the mechanisms driving perceptions of support (Reis and Collins, 2000).

Figure 2.5. Network features and individuals' predispositions determine relationship properties and interactions, which in turn drive perceptions of support, behavior, and ultimately health. Adapted from Reis and Collins (2000).
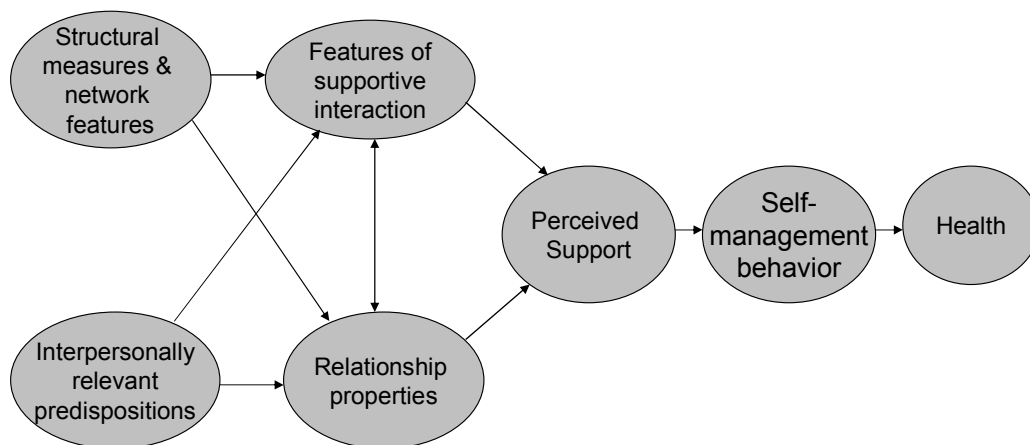
As briefly described above in the discussion of the correlates of self-management behavior, considerable empirical evidence from observational studies supports the notion that social support may be an important influence on self-management behavior in a variety of chronic illnesses. In separate, cross-disease, systematic reviews of observational studies, DiMatteo (2004) and Gallant (2003) concluded that social support is consistently and positively associated with self-management behavior across chronic illnesses.

In her meta-analysis of the relationship between social support and treatment adherence, DiMatteo (2004) found that functional measures of support (e.g., practical, emotional, and general support, family cohesiveness, and family conflict) were more strongly related to adherence than structural measures of support (e.g., marital status and living alone versus with others). All of the functional measures studied demonstrated significant relationships with treatment adherence: instrumental support, emotional support, general (uni-dimensional) support, and family cohesiveness were strongly and positively related to treatment adherence across studies, and family conflict was strongly and negatively related to adherence. The strongest relationship to treatment adherence was found for instrumental social support, compared to the other measures of functional support. Whether support was perceived to be available or actually received did not seem to matter with regard to treatment adherence; both had positive influences.

Gallant (2003) conducted a systematic review of the empirical literature that examined social support as a predictor of chronic illness self-management. Of the 13 studies she deemed to be of good methodological quality, 12 demonstrated significant, positive, and

small-to-moderate relationships between social support and at least one illness management behavior or type of treatment adherence. There was some evidence that illness- or regimen-specific support was more strongly related to adherence than general support, and that dietary and exercise behavior may be more influenced by support than other illness management behaviors (e.g., medication adherence). Gender and race were discussed as potential moderators of the relationship between support and adherence, but the number of studies examining these variables was too few to draw any firm conclusions. Gallant also pointed out that while qualitative investigations routinely found that friends and family members sometimes acted in unsupportive ways that made adherence more difficult, quantitative investigations of the processes by which social relationships negatively impact adherence were very rare.

While the results of both of the systemic reviews strongly suggest that we can view social support as an important influence on self-management behavior, it is important to note some limitations of the existing studies. First, the causal direction of the association between social support and self-management cannot be determined from these studies; good self-management could elicit more support from others than poor self-management, or a third variable could influence both support and self-management (DiMatteo 2004). If this is indeed the case, the true influence of support on self-management behavior may be smaller than suggested by the above reviews. There is also some suggestion that support may play a stronger role in determining behavior in some diseases versus others. Gallant's review found some evidence that support may matter more for diabetes than other illnesses, although she noted that the support-behavior relationship was studied more often in diabetes than any other illness. Additionally, DiMatteo did not find any cross-disease differences in her

quantitative review. In addition, it should be noted that problems with measuring social support and self-management behavior were noted in both of these reviews; for example, support was often defined vaguely and self-management behavior was most commonly measured using only self-report. However, while DiMatteo found that stronger relationships between support and adherence were found in studies measuring adherence via self-report, support remained related to adherence in studies using alternative methods of measurement.

It is also important to note that these reviews necessarily mixed different conceptualizations and measurements of support in order to draw conclusions about the general effects of social support on self-management behavior. For instance, neither author was able to investigate source of support as an important variable in their reviews because source was not often explicitly considered in observational studies (DiMatteo, 2004). However, the type of support expected and received by individuals may vary across different types of relationships, and support from romantic partners, friends, and family members may be differentially associated to various behaviors and health outcomes (Sarason, Sarason, et al., 1997).

The Role of Spouses

One potential source of support that may be particularly important with regard to self-management behavior among the chronically ill is support from close romantic partners such as spouses. Not only is marital status, regardless of its quality or level of support exchanged between spouses, related to treatment adherence (DiMatteo, 2004), but both quantitative (Glasgow and Toobert, 1988; Christensen, Smith, et al., 1992; Courneya, Blanchard, et al., 2001; Fraser, Hadjimichael, et al., 2003; Wen, Parchman, et al., 2004) and qualitative (Maclean, 1991; El-Kebbi, Bacha, et al., 1996; Trief, Sandberg, et al., 2003) studies have

implicated patients' experiences and perceptions of support from family members and the spouse in particular as having an effect on patients' ability to carry out their treatment regimens. These findings are in line with many researchers' assertions that close romantic partners such as spouses play a key role in determining overall perceptions of available support (Sarason, Sarason, et al., 1997; Reis and Collins, 2000) and enhancing treatment adherence and psychological adjustment among chronically ill patients (Revenson, 1993).

The studies that have linked spouse and/or family support to self-management behavior in adult patients with chronic illness are heterogeneous with regard to the illness population studied as well as their conceptualization of spouse support. However, a common thread among this literature is again the researchers' tendency to limit their investigations of spouse support and self-management behavior to establishing an independent association among perceptions of support and self-management without regard to the processes and mechanisms driving the relationship.

Many studies of self-management behavior in chronically ill patients have conceptualized social support in terms of patients' perceptions of the support available and received from spouses, or family members more generally.   These studies have often found positive associations between patients' perceptions of spousal/family support and treatment adherence. For example, in a study of adults with multiple sclerosis, adherent patients were more likely to report their spouse or their physician as the most supportive person of their therapy compared to non-adherent patients (Fraser, Hadjimichael, et al., 2003). In a study of Type II diabetics, higher levels of illness-specific received support from family members were significantly related to dietary, exercise, glucose-testing, and medication adherence (Glasgow and Toobert, 1988). Wen and colleagues (2004) similarly found that higher levels of diet-

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specific received family support was an independent predictor of perceived barriers to dietary adherence among Hispanic Type II diabetics, controlling for a variety of other influences on adherence. While these two studies did not separate spouse support from family support, family support was operationalized in terms of the individual with whom the patient reported interacting the most and thus was often likely the patient's spouse. In a study of exercise adherence among women with breast cancer, perceptions of available support from spouses was a significant predictor of intention to exercise and attendance at an exercise class, in addition to support from physicians and friends (Courneya, Blanchard, et al., 2001).

Other studies that have conceptualized spouse support in terms of marital relationship properties and interactions have also found significant associations with self-management behavior. While most studies adopting a relationship processes approach to studying social support and self-management behavior have focused more broadly on family relationships and not marital relationships specifically (e.g., see (Christensen, Smith, et al., 1992; Garay-Sevilla, Nava, et al., 1995; Wen, Parchman, et al., 2004), a few have linked marital relationships properties to self-management. In a cross-sectional analysis of heart disease patients and their spouses, marital functioning was significantly related to patients' adherence to diet, smoking and stress-reduction regimens, although the authors acknowledged that the causal relationship between martial functioning and adherence was likely bidirectional (Miller, Wikoff, et al., 1990). In a study of patients receiving dialysis treatment and their families, sharing of leadership and control by spouses significantly predicted patients' adherence to their treatment regimens (Steidl, Finkelstein, et al., 1980). However, multivariate analyses of this relationship were not explored. In a more recent study of adult patients with diabetes, marital intimacy and general marital adjustment were significantly

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related to dietary adherence and adherence to physician recommendations in cross-sectional analyses that controlled for variety of potentially confounding factors (Trief, Ploutz-Snyder, et al., 2004). While this finding did not hold up in longitudinal analyses of 2-year follow-up data (marital intimacy and adjustment at baseline were not related to changes in adherence), the long follow-up period and small sample size (N=42) may have been responsible for the non-significant finding. This study also did not explore whether marital intimacy and adjustment influenced adherence via patients' perceptions of spousal support, which would be predicted by the relationship processes perspective.

In addition to these quantitative studies of marital relationship processes and treatment adherence, a few qualitative studies also warrant mention. Several qualitative investigations with chronically ill individuals have implicated marital conflict and negatively-viewed supportive attempts from spouses as barriers to individuals' performance of self-management behaviors. These studies have asked adults with diabetes, asthma, and heart disease to discuss ways in which interactions with spouses negatively impact their ability to care for themselves. Patients in these studies reported that they reacted negatively to some of their spouses' attempts at providing support; for example, spouses were sometimes perceived as nagging (Clark and Nothwehr, 1997), overreacting to illness symptoms (Clark and Nothwehr, 1997), or treating them as invalids (Clark, Janz, et al., 1994). Patients also cited marital discord, due to both illness and non-illness related factors, as interfering with their ability to care for themselves (Maclean, 1991; Clark, Janz, et al., 1994; Handron and Leggett-Frazier, 1994; Clark and Nothwehr, 1997). While marital conflict and negatively-perceived supportive attempts from spouses have been found to be significant predictors of coping and psychological adjustment in chronically ill patients (Revenson, Schiaffino, et al., 1991;

Holahan, Moos, et al., 1997; Manne, Taylor, et al., 1997), and *family* conflict more generally is predictive of poor treatment adherence, no quantitative studies of the effect of *marital* conflict specifically on treatment adherence could be identified.

In summary, results from previous studies suggest that in addition to perceptions of spousal support, specific marital relationship properties and interactions may influence patients' self-management behavior, and that marital conflict in particular is a potentially important relationship property that exerts a negative influence on treatment adherence.

Evidence from Couples' Intervention Studies

Despite the consistent demonstration of social support from spouses specifically as a predictor of self-management behavior in observational studies, interventions for chronically ill individuals have largely failed to capitalize on this observed association. First, relatively few couples-focused interventions reported in the literature have targeted self-management behavior or treatment adherence specifically as an outcome. Instead, couples-focused interventions have much more often targeted the physical and emotional health of chronically ill individuals, intermediate psychological outcomes like self-efficacy and coping with aspects of the illness, or the health behavior of non-chronically ill individuals. Many of the articles reporting on these interventions have discussed the potential for the interventions to improve illness behaviors but curiously did not evaluate adherence as a specific outcome (e.g., Keefe, Caldwell, et al., 1999; Martire, Schulz, et al., 2003; Karlson, Liang, et al., 2004; van Lankveld, van Helmond ,et al., 2004). Second, couples-focused interventions that have attempted to marshal support from spouses have, for the most part, yielded disappointing results in terms of effectively improving perceptions of spousal support and improving outcomes (Cutrona and Cole, 2000).

The few couples-focused interventions that have specifically targeted health behavior among chronically ill patients have yielded mixed results with regard to both improving spouse support and health behaviors. For example, one randomized trial including rheumatoid arthritis patients and their significant others found that patients assigned to the spouse participation condition actually reported worse fatigue, decreased self-efficacy, and no improvements in exercise behavior relative to the patient-only condition (Riemsma, Taal, et al., 2003). The authors also noted that the partner-participation condition did not effectively improve spouse support as intended. Another trial that included heart disease patients and their spouses compared a spouse participation condition to a patient-alone condition and a control condition also failed to find beneficial effects of spouse participation on smoking, exercise behavior, blood pressure control and weight control (Dracup, Meleis, et al., 1984). Unfortunately, perceptions of support were not evaluated in this study and statistical power to detect differences across intervention conditions was low. One trial that did yield promising results was a family support intervention aimed at improving medication adherence, appointment-keeping, weight control, and blood pressure control among hypertensive patients (Morisky, DeMuth, et al., 1985). In this trial, patients identified the family member with whom they talked most to participate in the study. The family member was then educated about blood pressure control and the importance of adherence, identified ways in which they could help the patient adhere to the regimen, and made an explicit commitment to helping the patient adhere. The family support intervention was compared to patient counseling by a health educator and group education using a factorial design, and analyses showed that the family support intervention was responsible for improved behavioral and health outcomes compared to the other two interventions. However, this trial

did not assess whether increases in patients' perceptions of family support were responsible for the favorable effects of the family support intervention.

Partner support interventions to improve health behavior in non-chronically ill populations are much more common, but have produced equally mixed results. A recent Cochrane review of partner support interventions for smoking cessation analyzed trials that compared a partner-support condition to an identical intervention not including partners and found overall disappointing results: neither smoking abstinence nor individuals' perceptions of partner support were greater in partner support conditions versus comparison conditions (Park, Schultz, et al., 2004). Lichtenstein and colleagues (1986) similarly found that three separate attempts to improve partner support to achieve smoking cessation were unsuccessful and also generally failed to increase partner support and decrease negative spouse behaviors. Couples-focused interventions for weight-loss have generally yielded more positive short-term results (Black, Gleser, et al., 1990), but have not been able to produce sustained weight control effects. Also, it is unclear whether inclusion of a spouse in interventions affected weight loss through improvements in spousal support.

Several researchers have attempted to understand why consistent observational findings regarding the importance of spousal support have not translated into effective health behavior and chronic illness interventions. First, many intervention studies have failed to evaluate whether or not couples-focused interventions effectively alter patients' perceptions of spousal support (Black, Gleser, et al., 1990). Of the studies that did explore spousal support as a mediator, several have shown that it is difficult to alter perceptions of spousal support (Lichtenstein, Glasgow, et al., 1986; Park, Schultz, et al., 2004). Second, operationalization of spousal support in intervention studies has varied widely, from simply including spouses

in patient education sessions, to building spouse's coping skills and efficacy, to educating spouses on what behaviors are supportive, to teaching couples specific problem-solving and communication skills. Operationalizing the support condition by simply including spouses in education sessions is particularly problematic, in that it is unlikely to produce an effective "dose" of spousal support being delivered to patients assigned to that condition (DeVellis, Lewis, et al., 2003). The assumption that spouses assigned to participate in education sessions with patients will become more adept at providing support to patients without specific training in support and communication is tenuous. Spouses participating in education sessions may even be *perceived* by patients as more supportive due to their participation, but may not gain any actual skill in providing support that encourages good behavioral and psychological outcomes. Indeed, several researchers have noted that interventions focusing on specific interactions and communication patterns tend to produce favorable results, yet most interventions have not operationalized support in this way (Lichtenstein, Glasgow, et al. 1986; Lassner, 1991; Keefe, Caldwell, et al., 1999; Riemsma, Taal, et al., 2003). Keefe and colleagues (1999) found direct support for this observation in osteoarthritis patients. They found that the condition teaching couples communication and dyadic coping skills yielded favorable improvements at 12-month follow-up in self-efficacy, pain, psychological distress, and physical disability compared to a condition that consisted of traditional patient education with spouse participation.

From these intervention results, it appears that more research identifying the specific patterns of interaction and communication that underlie patients' perceptions of spousal support is warranted. Such research has the potential to inform the development of more

effective couples-focused interventions to improve self-management behavior in the chronically ill, as well as health behavior and adjustment to chronic illness more generally.

An Alternative Model for Studying Social Support

Interdependence Theory

The relationship science literature, and specifically work rooted in interdependence theory, is helpful in identifying specific interaction patterns that may affect outcomes in chronic illness such as treatment adherence. Interdependence theory (Kelley and Thibaut, 1978) provides a comprehensive model for understanding how individuals in relationships affect one another's beliefs, attitudes, emotions, and behaviors. Interdependence theory has not often been explicitly applied to health behavior, although its basic premise that interacting partners mutually influence each other underlies much health behavior change research and intervention (Lewis, DeVellis, et al., 2002).

Figure 2.6 (adapted from Lewis, DeVellis, and Sleath, 2002) depicts how interdependence in dyadic relationships such as marriage affects health behavior and health outcomes. Interdependence refers to the process by which individuals in a relationship mutually influence each other. In couples where one spouse is facing a chronic illness, each spouse's thoughts, feelings and behaviors relating to the illness are determined in part by his/her own characteristics (the parallel one-sided arrows), in part by his/her spouse's characteristics (the crossed one-sided arrows), and finally by the reciprocal influence of both spouses' interactions (the double-sided vertical arrow between spouses). This model implies that it is possible to break down the determinants of patients' illness-related thoughts, feelings, and behaviors into effects due to patients' own characteristics, their spouses' characteristics, and the interactions of patients and spouses. However, as described above, most research on

support in the context of chronic illness has treated social support as a characteristic, or

resource, of either the patient or the spouse and has failed to take into account how spouses

interact with each other to affect individual and relationship-level outcomes. Thus,

interdependence theory is a useful approach from which to study social support in chronic

illness, as it explicitly highlights that couples' interactions can determine patient (and spouse)

outcomes, such as treatment adherence.

Figure 2.6. Interdependence model of social influence and interpersonal communication.
Adapted from Lewis, DeVellis, & Sleath (2002).
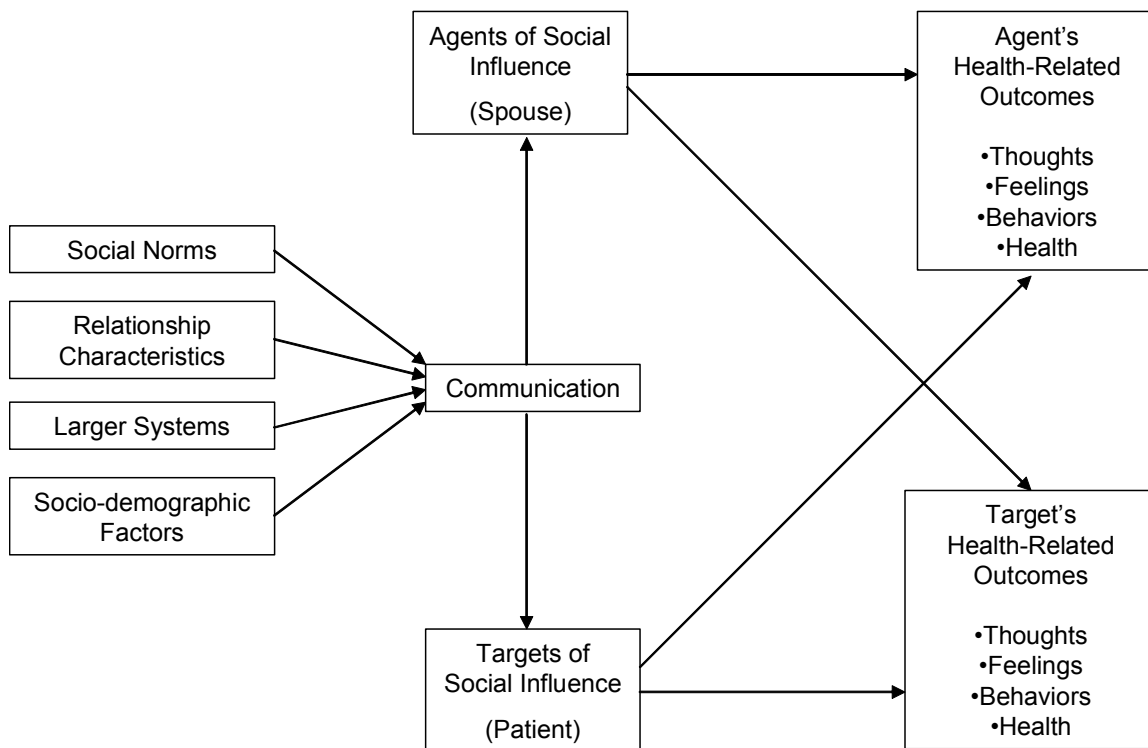
Transformation of Motivation and Accommodative Behavior

Two important constructs with origins in interdependence theory are transformation of

motivation and accommodative behavior. Interdependence theory suggests that as

commitment and interdependence in a relationship grows, partners tend to act in terms of what is good for the relationship and their partners in addition to what is good for themselves individually (Rusbult and Arriaga, 1997). Transformation of motivation refers to the process by which individuals (either consciously or automatically) prioritize their partners' and joint relationship goals and outcomes over their own immediate goals and outcomes (Rusbult and Arriaga, 1997). Figure 2.7 depicts this process of transformation of motivation. "Given matrix preferences" refers to individuals' "gut-level", primitive, and self-interested preferences for behavior, while "effective matrix preferences" are individuals' preferences for behavior that occur after taking into account feelings about one's partner and the relationship, long-term versus short-term relationship goals, one's interpersonal dispositions, and norms of the relationship. The process by which given matrix preferences are converted into effective matrix preferences and result in pro-relationship behavior is transformation of motivation. The model also shows the various factors that influence if, and how, self-interested preferences for behavior are transformed into pro-relationship preferences and behavior.

Figure 2.7. The process of transformation of motivation, from Rusbut & Arriaga (1997).
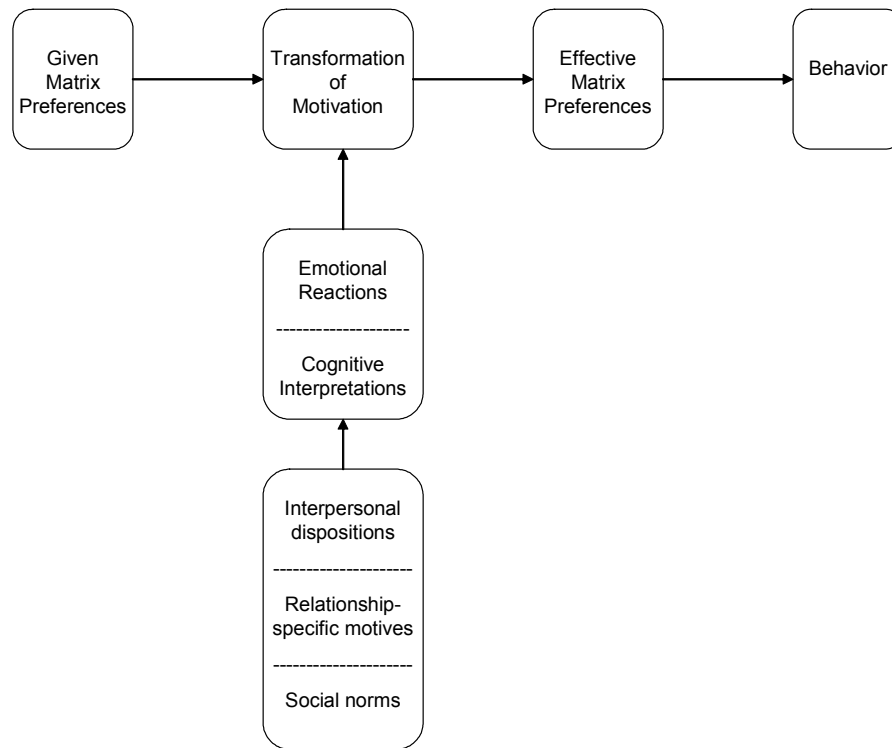
One indicator of the extent to which transformation of motivation has occurred for an individual in a particular relationship is the individual's tendency toward accommodation (Rusbult, Verette, et al., 1991; Wieselquist, Rusbult, et al., 1999). Accommodation is defined "as an individuals' willingness, when a partner has engaged in a potentially destructive behavior, to (a) inhibit tendencies to react destructively in turn and (b) instead engage in constructive reactions" (Rusbult, Verette, et al., 1991). In other words, accommodation refers to individuals' tendency to respond constructively rather than destructively to the inevitable conflict and dissatisfaction with behavior by partners that occur in all close relationships. Theory and empirical evidence suggests that individuals' level of commitment to the relationship is the most proximal determinant of accommodative behavior (Rusbult, Verette, et al., 1991; Rusbult, Bissonnette, et al., 1998).

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Theory and empirical work also suggests that accommodation is a multi-dimensional construct. Individuals' responses to destructive acts by their partners can be constructive or destructive in terms of their effects on the relationship, as well as active or passive with regard to their effects on the problem at hand. Rusbult and colleagues (1991) have defined four patterns of accommodative behavior: voice, loyalty, exit, and neglect. Voice is characterized by active, constructive attempts to resolve the situation (e.g., discussing problems and suggesting solutions). Loyalty is characterized by a passive, constructive response in which the individual passively expects and waits for things to improve (e.g., simply waiting until the partner's mood and behavior improves). Exit is an active, destructive response (e.g., yelling, threatening to leave, physical abuse), while neglect is a passive, destructive response (e.g., ignoring the partner, criticizing the partner for issues unrelated to the apparent conflict). Figure 2.8 depicts this typology of accommodative behavior. Voice and loyalty are together considered to be "constructive accommodation" while exit and neglect are considered to be "retaliation" (Rusbult, Bissonnette, et al., 1998).

Figure 2.8. Typology of accommodative behavior (Rusbult and Verette 1991).
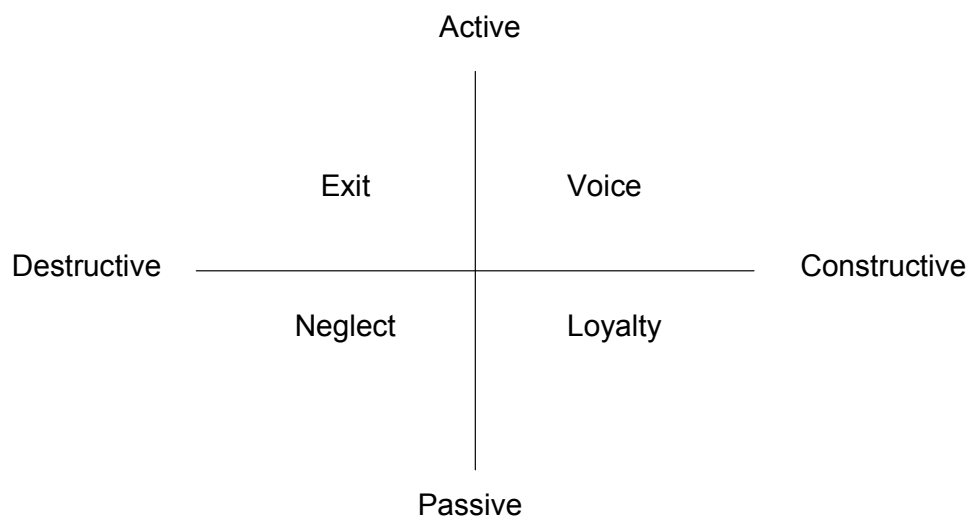
Accommodative Behavior and Relationship Outcomes

Although accommodation has not previously been studied in the context of chronic illness, several studies have been conducted to specify the impact of accommodative behavior on couples' adjustment, distress, and functioning. Like a lot of social psychological studies, these studies have primarily been conducted with healthy young couples in dating relationships or in the early years of marriage and therefore may not have produced results that are generalizable to older couples who have been married for many years or who are facing a long-term illness like vasculitis. However, this body of research has shown good evidence that couples' accommodative behavior, and particularly one's tendency to retaliate, or respond to conflict in destructive ways, influences couple functioning (Rusbult, Johnson, et al., 1986; Rusbult, Verette, et al., 1991; Wieselquist, Rusbult, et al., 1999).

Rusbult and colleagues (1986) demonstrated in cross-sectional analyses that self-reported retaliation was positively correlated with couples' distress, and self-reported constructive accommodation was negatively correlated with couples' distress. In another cross-sectional study, Rusbult and colleagues (1991) demonstrated that couple distress was highest when both partners in a relationship reported greater tendencies toward retaliation. However, the cross-sectional designs of these studies limited the researchers' ability to make strong arguments regarding the causal direction of the association between couples' retaliation and distress. That is, these studies were not able to conclude with any confidence whether retaliation causes relationship distress or if relationships distress causes retaliation, and it is very possible that the relationship between accommodation patterns and distress is reciprocal as relationships progress over time.

Two more recent longitudinal studies have provided more convincing evidence that accommodation is a determinant of couple functioning and distress. In studies of young married couples (Rusbult, Bissonnette, et al., 1998; Wieselquist, Rusbult, et al., 1999) and young adults in dating relationships (Wieselquist, Rusbult, et al., 1999), self-reported accommodation (as measured using a composite of both constructive and destructive responses to conflict) and perceptions of partners' accommodation were both significantly and positively associated with concurrent levels and prospective changes in levels of trust and dyadic adjustment. These studies also demonstrated that both constructive accommodation and retaliation independently affected dyadic adjustment, although effects of retaliation were larger (Rusbult, Bissonnette, et al., 1998), and that perceptions of spouses' accommodation more powerfully predicted dyadic adjustment than individuals' own reports of accommodation (Rusbult, Bissonnette, et al., 1998). There is also some evidence that the strength of the association between accommodation and dyadic adjustment increases over time as the marriage progresses (Rusbult, Bissonnette, et al., 1998). Finally, these studies also explored the possibility of gender differences and concluded that there is little evidence that the influence of accommodative behavior on dyadic adjustment varies by gender (Wieselquist, Rusbult, et al., 1999).

Accommodation: A Possible Determinant of Perceptions of Support and Illness Self-Management?

Rationale

The relatively limited body of work on the consequences of partners' accommodative behavior in marital relationships provides a fairly convincing argument that accommodation may act as an important determinant of couple functioning and adjustment. If individuals'

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responses to relationship conflict and dissatisfaction with partners determine dyadic adjustment, then it is not unreasonable to hypothesize that accommodation in marital relationships may affect individuals' perceptions of support from partners. If accommodation is a mechanism underlying perceptions of spouse support, then outcomes specific to the context of chronic illness, such as patients' self-management behavior, could also be affected by accommodation.

The evidence from qualitative and quantitative studies implicating marital conflict as a relevant determinant of self-management behavior further suggests that patterns of accommodation in marital relationships may be relevant to illness self-management. As discussed above, qualitative studies suggest that patients perceive negative interactions with spouses as interfering with their ability to carry out illness management tasks and quantitative studies have found significant negative associations between family conflict and treatment adherence. However, the practical implications of these studies for intervention development are somewhat limited, as some instances of conflict and dissatisfaction with partners in marital relationships are inevitable, particularly for couples facing the stress and role changes that life with chronic illness often brings. In contrast, studying how partners *respond* to conflict and dissatisfaction as a determinant of self-management behavior could potentially identify specific patterns of interactions to target in future interventions for couples facing a chronic illness.

Conceptual Model for Manuscript #3

Figure 2.9 below depicts a conceptual model of how accommodation in marital relationships is hypothesized to affect patients' self-management behavior. The model combines ideas from the traditional "direct effects" model of the influence of social support

on health behavior, in which perceived support directly influences health behavior (Burman & Margolin, 1992), and research drawing on Interdependence Theory, in which accommodative behavior on behalf of both spouses influences perceptions of one's relationship functioning. Considering the model from left to right, spouses' accommodative behavior and patients' own accommodative behavior are hypothesized to be correlated with one another (as depicted by the double-headed curved arrows). Paths are then drawn leading from spouse retaliation and constructive accommodation and patient retaliation and constructive accommodation to patients' perceptions of spousal support. This reflects the hypothesis that perceptions of spousal support are jointly determined by each spouse's accommodative behavior. Finally, patient perceptions of spousal support are then shown to directly influence patients' self-management behavior.

Possible alternative models. It should be noted that the lack of arrows leading directly from the accommodation variables to self-management behavior implies that accommodation in marital relationships exerts its effect on health behavior entirely through patient perceptions of spousal support. It is possible that spouse accommodation directly influence patients' self-management or operate through pathways other than social support. In manuscript #3, this possibility is explored.

Figure 2.9. Relationship processes model of accommodation, spousal support, and treatment adherence.
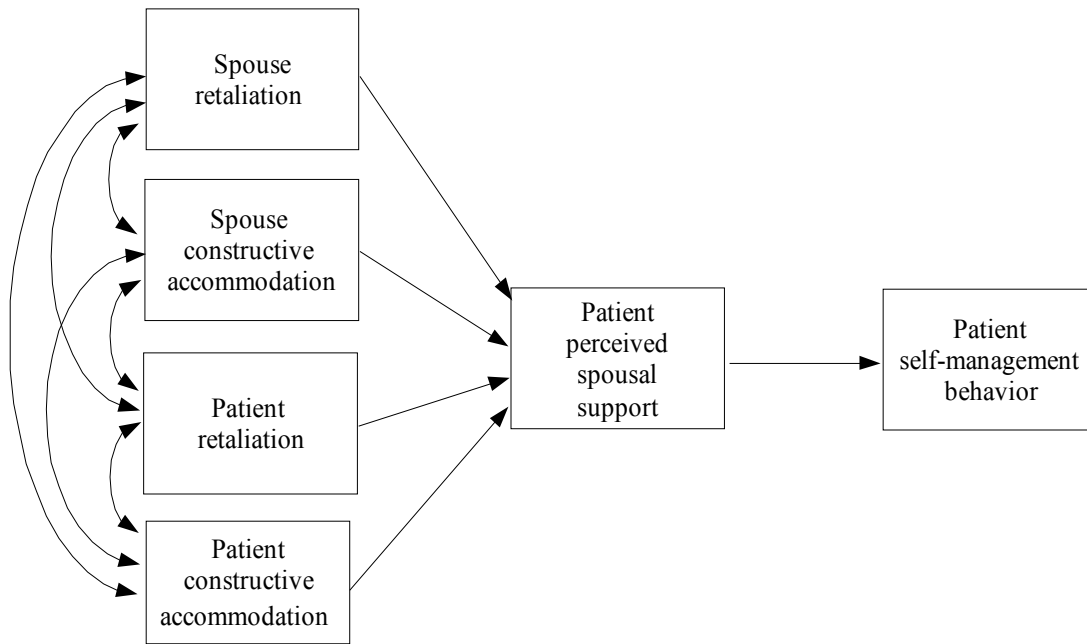
CHAPTER THREE: RESEARCH AIMS AND HYPOTHESES

Based on the literature described above and formative work conducted with ANCA-SVV patients (described more in Chapter 4), the following specific aims, research questions, and hypotheses formed the basis of the dissertation. While specific hypotheses were tested in manuscripts #1 and #3, the exploratory and descriptive nature of manuscript #2 led me to examine broader research questions rather than test specific hypotheses.

Specific Aim #1 (Manuscript #1): To develop a self-report measure of ANCA-SVV self-management behavior and gather evidence of its reliability and validity.

H1: Self-management behavior among ANCA-SVV patients is multi-dimensional; that is, multiple, independent domains of illness self-management behavior underlie the new measure of ANCA-SVV self-management behavior.

H2: Domains of self-management behavior will be only modestly correlated with one another.

H3: Each subscale of the self-management measure will demonstrate acceptable internal consistency reliability (i.e., Cronbach's $\acute{a}$ greater than .70) and test-retest reliability.

H4: Correlations between scores on the new self-management measure and scores of other indicators previously shown to have relevance to self-management behavior in other chronically ill populations will be statistically significant and of a magnitude and pattern consistent with construct validity. Table 3.1 below shows the specific predicted relationships.

Table 3.1. Direction and magnitude of hypothesized relationships between validation

measures and vasculitis-specific self-management behavior.

Variable	Predicted direction of association	Predicted magnitude of association
Socio-demographics		
Age	Zero	Zero
Gender	Zero	Zero
Education	Positive	Small
Clinical and health variables		
Duration of diagnosis	Negative	Small to moderate
Current disease activity	Positive	Small to moderate
Disease damage	Positive	Small to moderate
Regimen characteristics		
Perceived treatment complexity	Negative	Small to moderate
Number of prescribed medications	Negative	Small to moderate
Number of recommendations	Negative	Small to moderate
Psychosocial and behavioral characteristics		
Social desirability	Zero	Zero
General adherence to medical recommendations	Positive	Small
Perceptions of available social support	Positive	Small to moderate
Vasculitis-specific self-efficacy	Positive	Small to moderate
Depressive symptoms	Negative	Moderate

Specific Aim #2: To characterize patient attitudes and beliefs, including perceived barriers

and facilitators, about performing self-management behaviors among adults living with

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ANCA-associated small vessel vasculitis (ANCA-SVV), and determine if these attitudes and beliefs were associated with levels of self-management behavior.

Q2a: Which self-management behaviors do patients generally find most difficult to perform?

Q2b: Which behaviors do they believe are the most important to perform?

Q2c: What are the specific factors that patients perceive as impeding or facilitating the performance of these behaviors?

Q2d: Do patients' perceived difficulty and importance, as well as their reporting of specific barriers and facilitators, predict levels of self-management behavior?

Specific Aim #3: To investigate spousal accommodative behavior as a supportive process that facilitates patients' chronic illness self-management behavior.

H1: Higher levels of retaliation by spouses are negatively associated with patients' perceived spousal support, independent of the spouse's level of constructive accommodation and the patient's own accommodative behavior.

H2: Higher levels of constructive accommodation by spouses are positively associated with patients' perceived spousal support, independent of the spouse's level of retaliation and the patient's own accommodative behavior.

H3: Patients' perceived spousal support is positively associated with their level of self-management behavior.

H4: Patients' perceived spousal support completely mediates the relationship between spousal accommodative behavior and patients' self-management behavior.

CHAPTER FOUR: DEVELOPMENT AND INITIAL EVALUATION OF A MEASURE OF SELF-MANAGEMENT FOR ADULTS LIVING WITH ANCA-ASSOCIATED SMALL VESSEL VASCULITIS

Abstract

Objective: To develop a measure of illness self-management appropriate for adults diagnosed with ANCA-associated small vessel vasculitis (ANCA-SVV) and to gather evidence of the measure's reliability and validity.

Methods: Development of the Vasculitis Self-Management Scale (VSMS) was guided by previous research on self-care behavior in other chronically ill populations, a review of the current treatment literature for ANCA-SVV, interviews with patients, and consultation with experts in ANCA-SVV, self-management, and scale development. A total of 205 patients living with ANCA-SVV or a closely related condition completed the VSMS, along with measures of a variety of socio-demographic, clinical, regimen, psychosocial, and behavioral variables, using a self-administered questionnaire. An exploratory factor analysis was conducted on the 68 self-care items. Internal consistency reliability (as indicated by Cronbach's $\acute{\alpha}$) and construct validity of the resulting subscales were assessed. Forty-five patients then completed the VSMS a second time for the purpose of calculating test-retest correlations.

Results: Analyses suggested an eight-factor solution for the VSMS. The final scale consisted of 44 items representing eight behavioral domains. Correlations among the eight factors underlying the measure were null to modest in magnitude. The internal consistency reliability of the eight subscales ranged from minimally acceptable ($\acute{\alpha}$=.67) to excellent ($\acute{\alpha}$=.94), and correlations between subscale scores at Time 1 and Time 2 suggested good temporal stability of the measure. Evidence for construct validity was mixed.

Conclusions: These findings suggest that the VSMS is a promising method for assessing illness self-management in adults living with ANCA-SVV. More research exploring the validity of the measure is warranted.

Introduction

ANCA-associated small vessel vasculitis (ANCA-SVV) is a group of rare autoimmune conditions, characterized by inflammation and necrosis of blood vessels primarily in the respiratory tract and kidneys (Watts & Scott, 1997), with an overall incidence estimated around 39 per million (Watts *et al.*, 1995). ANCA-SVV is a potentially fatal, relapsing illness, with an often progressive but unpredictable course (Koutantji *et al.*, 2000). Until recently, rapid death from ANCA-SVV was likely (Fauci *et al.*, 1983), but the introduction of effective immunosuppressive medications has transformed ANCA-SVV into a chronic illness with which most patients live for many years (Koutantji *et al.*, 2000). There is no cure for the illness, but treatment with immunosuppressive medications induces remission in 80-100% of patients (Langford, 2003). Patients may experience full remission, partial remission, and/or minor or major relapses (Jayne, 2003). Even among patients in remission, the risk of relapse and morbidity from the disease and its ongoing, highly toxic treatment remains high (Hoffman *et al.*, 1998).

As with other serious chronic illnesses, individuals who are diagnosed with ANCA-SVV face a daunting array of psychosocial challenges. In addition to living with an unpredictable and potentially fatal prognosis, patients and their families are asked to undertake complicated treatment regimens that come with considerable risk of serious side effects (Jayne, 2003) and require substantial role adjustment and lifestyle changes (Koutantji *et al.*, 2000). Yet leading researchers have noted the paucity of research on the psychosocial aspects of living with vasculitis. In an editorial outlining some of the unanswered questions about the psychological aspects of vasculitis, Koutantji, Pearce, and Harrold (2000) suggested that a

major goal of future research should be to evaluate coping, functioning, and quality of life issues in individuals living with vasculitis.

One of these issues not previously studied is patients' illness self-management, which is defined as the "day-to-day tasks an individual must undertake to control or reduce the impact of disease on physical health status" (Clark *et al.*, 1991). Patients assume these tasks with the collaboration and guidance of their health care providers (Clark *et al.*, 1991). A construct related to illness self-management is treatment adherence, or "the extent to which a person's behavior – taking medication, following a diet, and/or executing lifestyle changes – corresponds with agreed-upon recommendations from a health care provider" (WHO, 2003). Although many self-management tasks include adhering to physicians' recommendations, self-management is a broader construct of which treatment adherence is one aspect (Gallant, 2003). Self-management involves mastery of three types of activities: performing activities to manage the condition, making informed decisions about care, and applying skills to maintain adequate psychosocial functioning (Clark *et al.*, 1991). Specific self-management behaviors required by patients vary across conditions (Clark *et al.*, 1991).

Self-management of ANCA-SVV has very rarely been researched; however, a review of the ANCA-SVV treatment literature suggests that self-management in ANCA-SVV typically requires patients to take medications, obtain vaccinations, regularly use health services, monitor symptoms of disease activity and treatment side effects, communicate with physicians, and/or change other aspects of their lifestyles (WGAI, 2003). Specific behavioral regimens are often adjusted based on current disease activity, side effects, and complications (WGAI, 2003). Patients usually remain on these regimens for many years, even after remission is attained (Hoffman *et al.*, 1998).

Next to nothing is known about the extent to which ANCA-SVV patients actually perform these self-care behaviors; however, patients' self-management of other chronic illnesses is often less than ideal. For example, typically only 50% of patients, regardless of the specific illness, adhere to their doctor-recommended treatment regimens (WHO, 2003). Poor self-management in ANCA-SVV could potentially be very problematic, in that following self-care regimens "is a primary determinant of the effectiveness of treatment" (WHO, 2003). Indeed, Andrassy and colleagues (Andrassy *et al.*, 1991) found that poor medication adherence was responsible for renal failure in two of 25 ANCA-SVV patients followed for a three-year period. Failure to carry out ANCA-SVV self-management tasks could lead to illness complications, impaired quality of life, or death.

Complicating the study of illness self-management and adherence in these patients is the lack of an existing self-report measure that can reliably, validly, and conveniently measure ANCA-SVV patients' performance of relevant self-care behaviors. Development of such a measure would allow for the empirical study of important influences on self-care behaviors in this population, which would in turn provide guidance for interventions that are tailored specifically for vasculitis patients to improve their performance of these behaviors.

Thus, the first goal of this study was to develop such a measure of ANCA-SVV self-management. ANCA-SVV self-management was defined in this study as performance of behaviors directly recommended by health professionals to patients for controlling the condition and maximizing physical and psychosocial functioning, and also performance of self-care behaviors generally recommended for all ANCA-SVV patients. Thus, we aimed to develop a measure that tapped into both treatment adherence, traditionally defined as the frequency with which patients' behavior corresponds with medical recommendations (WHO,

2003), as well as performance of other relevant self-care behaviors, such as appropriate responding to illness symptoms, that are important in self-management of ANCA-SVV. Additionally, we aimed to develop a self-report measure that is flexible for use with a majority of patients living with vasculitis, regardless of their disease activity or specific treatment recommendations, but that also provides as much specific information as possible about patients' adherence to different behavioral recommendations. In addition, we sought to develop a measure that allowed for multiple domains of adherence behavior, given that prior research has shown that rates of non-adherence vary across different self-care behaviors (Johnson, 1992; Toobert & Glasgow, 1994; WHO, 2003). For example, research with adult diabetes patients has repeatedly shown that adherence to one component of the diabetes regimen is not highly correlated with adherence to other components of the regimen (Johnson, 1992; Toobert & Glasgow, 1994). Thus, we hypothesized that multiple, independent domains of illness self-management behavior would underlie the newly developed measure. Further, we expected these behavioral domains to be only modestly correlated with one another, if at all.

The second goal of this study was to gather evidence of the new measure's reliability and validity. Specifically, we expected each subscale of the self-management measure to demonstrate internal consistency reliability (measured by Cronbach's α) greater than .70, the cutoff widely considered to be acceptable for between-group comparisons (DeVellis, 2003). Furthermore, we expected adequate test-retest reliability, or high within-subscale correlations when the measure was administered to the same participants at two points close in time. Finally, we expected correlations between scores on the new self-management measure and scores on measures of other constructs previously shown to be related to self-care behavior in

other chronically ill populations to be statistically significant and of a magnitude and pattern consistent with construct validity. Specific predicted relationships are shown in Table 4.1.

Patients and Methods

The Vasculitis Self-Management Scale (VSMS) was developed in four main steps: 1) definition of the construct (described above) and identification of the specific behaviors to be measured, 2) initial item generation and questionnaire design, 3) pilot-testing, and 4) scale administration, item and dimensionality analysis, and reliability and validity testing.

Identification of Behaviors and Initial Scale Development

Identification of the specific behaviors to be assessed by this measure and subsequent item generation was based on a review of the literature as well as semi-structured, in-depth telephone interviews with 18 adult patients living with ANCA-SVV. Potential domains of self-care behavior were identified through a review of the literature on current treatment for ANCA-SVV (Hoffman & Drucker, 1998; Jayne, 2003; WGAI, 2003). In subsequent interviews, patients were asked to indicate which of the behaviors identified in the literature review had been recommended to them by their health professionals, and to describe in detail these recommendations, along with the barriers and facilitators they experienced in performing them.

Through these methods, 11 different behaviors (8 adherence behaviors and 3 general self-care behaviors) were identified that were hypothesized to apply to a majority of individuals living with vasculitis: 1) adherence to prescribed medications taken at home, 2) adherence to prescribed medication administered by a health professional, 3) adherence to recommended appointments with health professionals, 4) adherence to recommended medical tests and immunizations, 5) adherence to recommended infection avoidance behaviors, 6) dietary

adherence, 7) exercise adherence, 8) adherence to symptom monitoring recommendations, 9) adjusting one's activity level based on current symptoms and fatigue, 10) prompt reporting of new or increased illness symptoms to a health professional, and 11) prompt reporting of new or increased treatment side effects to a health professional. Because the number of possible behaviors central to vasculitis self-management is quite large and variable across patients, we decided to exclude from the measure those behaviors that are not commonly required of most ANCA-SVV patients (e.g., smoking cessation). Items assessing performance of each of 11 self-care behaviors were then written, with guidance from phraseology used in other well-established self-report measures of self-care and treatment adherence (e.g., the Summary of Diabetes Self-Care Activities; Toobert & Glasgow, 1994) and the General Adherence Scale; Hays, 1993; Kravitz *et al.*, 1993)).

Pilot-testing and Measure Revision

In the second phase of the research, both professionals and patients provided feedback on the content, clarity, specificity, and wording of the draft VSMS. First, nine researchers and clinical experts in the fields of health behavior, illness self-management, scale development, and/or ANCA-SVV provided feedback on the items, which informed the first set of revisions. The revisions at this stage were minor and largely consisted of wording changes and added instructions so that the measure would be appropriate for a broader range of ANCA-SVV patients (e.g., those in remission with a high level of functioning, as well as those with substantial morbidity and functional limitations). The revised adherence measure was then pilot-tested with eight vasculitis patients. Patients completed the draft measure and then provided feedback via semi-structured telephone interviews. This feedback informed further

revisions of the measure, which included minor wording and formatting changes and the

addition of instructions to improve clarity.

The final version of the VSMS administered to the large patient sample consisted of 68

items assessing patients' performance of the 11 behaviors described above (the full VSMS is

available from the first author upon request). Within each of the eight adherence sections,

patients were first asked if a health professional had recommended that they perform the

particular type of behavior (e.g., taking medication), and then to provide more details about

the specific behavioral recommendations (e.g., how many, when, and how medications were

supposed to be taken). If patients had not been asked to perform the type of behavior, they

were instructed to skip to the next section of the questionnaire. Patients who had been asked

to perform the behavior were then asked follow-up questions about how frequently they

followed the recommendation(s) in recent weeks and/or months. Two different recall periods

were used, depending on the frequency with which patients are typically required to perform

the behavior. For behaviors that are typically performed on a daily basis (e.g., medication-

taking, following dietary guidelines), patients were asked to report the frequency with which

they performed the behavior according to their physicians' recommendations in the past 4

weeks. This recall period was thought to be long enough to produce sufficient opportunity

for non-adherence and, in turn, variation in responses among respondents, but short enough

to produce reliable and accurate self-reports of behavior. For behaviors that are performed

with varying frequency and/or much less often than daily (e.g., attending appointments)

patients were asked to report their behavior during the past year. The response format varied

across items, but consistently used five-point Likert scales (e.g., *none of the time* to *all of the*

time). Several items pertaining to each self-care behavior were negatively worded and

reverse-scored, so that higher scores on each item corresponded to higher levels of adherence. The format of the sections that assessed the remaining three self-care behaviors (adjusting activity level in response to symptoms, reporting illness symptoms, and reporting side effects) was similar to the format of the adherence section. The exception was that patients only skipped these sections if they had not experienced any symptoms during the recall period. That is, the skip pattern was not based on their physicians' recommendations because our conversations with ANCA-SVV health professionals and patients revealed that these self-care behaviors are appropriate for all ANCA-SVV patients when they experience fatigue or illness symptoms.

Administration and Analysis of the Final Measure

Participants and Procedures

To be eligible for the study, patients were required to be 18 years or older, be able to read and write in English, and have been diagnosed with a form of ANCA-SVV or a closely related condition. Autoimmune conditions that are generally treated using the same types of medication, tend to affect the same organs, and have similar prognoses as ANCA-SVV (e.g., Good Pasture's disease) were included. Adult patients with these conditions were recruited for participation by posting announcements about the study in a variety of places. First, patients 18 years or older who were part of an ANCA-SVV and glomerulonephritis patient disease registry at the lead author's institution and had signed long-term consent to be contacted about vasculitis research were invited to participate via letter. Announcements about the study were also placed in newsletters targeted toward ANCA-SVV patients and on several ANCA-SVV-related websites. Finally, ANCA-SVV patients who had volunteered for another study in which the lead author was involved were asked if they were interested in

hearing about other ANCA-SVV research, and if they responded positively, they were invited to participate in this study. A total of 278 interested patients were identified through these methods and mailed study consent forms. Of these, 233 (84%) signed and returned the consent form, and 208 (75%) completed and returned the study questionnaire. Two participants were subsequently excluded from the analyses because they or their doctor reported a primary diagnosis of lupus and one participant was excluded because he reported an unknown diagnosis which his physician later indicated was not vasculitis-related. This resulted in a final sample of 205 patients who completed the self-management measure along with the measures described below. In addition, 48 participants were randomly selected and asked to complete the self-management questionnaire a second time, approximately four weeks following the initial administration. Forty-four (92%) returned completed Time 2 questionnaires an average of 6.2 weeks (SD=2.7 weeks) after their Time 1 questionnaires (range = 3.4-15 weeks). Participants were provided with a $10 gift (in the form of cash or a gift card) for each questionnaire they completed and returned.

Measures

  Socio-demographics. Patients were asked to provide basic socio-demographic information, including age, gender, race/ethnicity, education, and marital status.

  Clinical and health variables. Patients were asked to report any other major medical diagnoses and their history, if any, of dialysis and kidney transplant. Illness duration, in months, was assessed by asking participants to report the month and year of diagnosis with ANCA-SVV, and then subtracting these values from the month and year of questionnaire completion. Current disease activity was assessed by both physician and patient report. Patients were asked to rate the current activity of their vasculitis on a scale from 1 (not active

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at all/remission) to 10 (extremely active). Physicians were also asked to use the same rating scale to assess the patient's disease activity. Morbidity and damage from vasculitis were also assessed via physician report using a similar 10-point scale (1=no damage to 10=extreme damage). For the purpose of characterizing the sample, current health status was assessed using the General Health Scale of the SF-36, Version 2.0, scored using the RAND method (J. E. Ware, Jr. & Sherbourne, 1992). Cronbach's *ά*, a measure of internal consistency reliability, was .82.

Regimen characteristics. Three indicators of treatment regimen complexity were collected: number of regimen components individuals report being asked to currently follow, number of medications patients report are currently prescribed for reasons related to their vasculitis or its complications, and patient-perceived treatment complexity. The latter indicator consisted of patient responses to the question, "In your opinion, how complicated is your treatment plan?" The response format consisted of a 10-point Likert-scale (range from 1 to 10) with higher ratings corresponding to greater perceived treatment complexity.

Psychosocial and behavioral variables. A number of psychosocial and behavioral variables were also assessed: social desirability, general adherence, social support, self-efficacy, and depressive symptoms.

Social desirability, or the desire of respondents to appear in ways that society regards as positive, was assessed using the short form of the Marlowe-Crowne Social Desirability Scale (Strahan & Gerbasi, 1972). This scale is widely used in psychometric studies to determine whether answers to self-reported questionnaires are influenced by respondents' desires to appear in socially desired ways. Cronbach's *ά* in this study was .77.

Patients' general tendency to adhere to medical recommendations was measured using the five-item General Adherence Scale from the Medical Outcomes Study (Hays, 1993). The General Adherence Scale does not measure adherence to specific self-care recommendations, but rather, patients' general tendency to adhere to medical recommendations made in the past four weeks. The initial testing of the General Adherence Scale demonstrated that it has adequate internal consistency reliability ($ά=.81$) and moderate test-retest reliability ($r=.40$; Hays, 1993). Cronbach's $ά$ in this study demonstrated adequate internal consistency reliability ($ά=.70$). Scores on the General Adherence Scale have been previously shown to have only a small correlation with scores on behavior-specific measures (Hays, 1993) and show only a very small correlation with scales measuring social desirability (Hays, 1993). Additionally, the General Adherence Scale has demonstrated construct validity in a sample of diabetics, in that higher general adherence was correlated with fewer hyperglycemic symptoms, lower blood glucose, and lower body mass index (Kravitz et al., 1993).

Patients' perception of the availability of support from others was assessed using the Social Provisions Scale (Cutrona & Russell, 1987). This measure is designed to assess six supportive functions provided by social network members: reliable alliance (belief that others can be relied on for tangible assistance), attachment (emotional closeness and a sense of security provided by others), guidance (advice or information available from others), nurturance (the sense that others rely on the patient for their well-being), social integration (a sense of shared interests and concerns with others), and reassurance of worth (others' recognition of the patient's competence, skills, and value). Previous research has shown that higher overall scores on the Social Provisions Scale predict better treatment adherence among end-stage renal disease patients (Moran *et al.*, 1997) and higher scores on the

attachment subscale predict greater medication adherence among individuals with HIV (Catz *et al.*, 2000). Based on this previous research and reports that the overall scale shows considerably higher reliability than the individual subscales (Wills & Shinar, 2000), we used the overall score on the Social Provisions Scale for our analysis (*ά*=.88).

Vasculitis-specific self-efficacy was assessed using a modified version of Lorig's arthritis self-efficacy scale (Lorig *et al.*, 1989). The original scale measures patients' confidence that they can perform behaviors to help them control their arthritis. The wording of the original scale was altered slightly to apply to vasculitis. When used with arthritis patients, the scale has demonstrated both excellent internal consistency reliability, with alphas above .90, as well as construct and concurrent validity (Lorig, et al., 1989). Cronbach's *ά* in this study was .89.

Patients' current level of depressive symptoms was assessed using the Center for Epidemiologic Studies Depression scale, or CES-D (Radloff, 1977). The CES-D is a continuous measure of depressive symptoms and has been used with a variety of populations, including individuals with various forms of chronic illness, and demonstrates very good internal consistency reliability, test-retest reliability, and various types of validity (Blalock *et al.*, 1989; Eaton *et al.*, in press). Cronbach's *ά* in this study was .88.

Analysis

All analyses were conducted using SAS Version 9.13 (SAS Institute, Inc., Cary, NC). Descriptive statistics for all variables described above, including the frequency (*n*), mean (*M*), and/or standard deviation (*SD*) and the 68 self-management items (*n*, distribution of responses, inter-item correlations) were first examined.

Handling of missing data. It was anticipated that all items would have some proportion of data missing, because not all self-management behaviors were expected to be relevant for all patients and as a result, patients would, as instructed, skip some items when completing the questionnaire. Because the goal of the study was to develop a measure of self-care behavior applicable for most ANCA-SVV patients, we specified *a priori* that items must have been applicable for and completed by over 50% of patients to include in the factor analysis. It was discovered that items regarding adherence to medications administered outside of the home by a health professional only applied to 25% (n=52) of the sample; thus, the six items referring to this self-care behavior were excluded from further analyses. The remaining 62 items were completed by a minimum of 60% (n=123) of participants, and were thus retained in subsequent analyses. Over half of the items were completed by over 85% of the sample, and the items for which data were missing most frequently pertained to exercise (39% missing) and symptom monitoring (35% missing).

It was assumed that missing data due to the skip patterns were missing at random (MAR), and therefore, ignorable. MAR assumes that the probability that data for a given variable are missing does not depend on the value of the variable itself, after other observed variables are controlled, although there is no test to determine whether this is the case (Allison, 2003). However, we assumed that the vast majority of missing data on the self-care items was not related to the individual's level of self-care, but instead was directly the result of the behavior not being relevant to the individual's specific illness situation, resulting in data that are MAR. When data are MAR and/or ignorable, a variety of techniques can be used when conducting factor analysis: listwise deletion, pairwise deletion, imputation of missing values, and full-information maximum likelihood (FIML) (Allison, 1987; Muthen *et al.*, 1987), with the latter

method being preferred (Allison, 2003) because of its improved accuracy in estimating variances and covariances. However, maximum likelihood estimation relies on the assumption that data are continuous and normally distributed, which can pose problems when conducting factor analysis, particularly when the sample size is small and the number of items is large (Flora & Curran, 2004). Indeed, when we attempted to run the factor analysis using FIML, estimates did not converge. Thus, we chose to use the pairwise correlation matrix in the factor analysis. This approach assumes, in essence, that the correlation between any two items obtained from the reduced sample is an acceptable estimate of the true correlation between those items.

Factor analysis. To determine the number of dimensions of self-care behavior that underlie the measure and to identify the items that measure those dimensions, a principal components analysis[1] (PCA) of the 62 items was conducted. Two complimentary methods were used to determine the number of common factors to extract: Cattell's scree test (Cattell, 1966) and parallel analysis (Horn, 1965). Like other widely used methods for determining the optimal number of factors underlying a set of items, both methods use eigenvalues, or the amount of information in the set of items that is captured by a factor (DeVellis, 2003). Based on their favorable performance in simulation studies, using these two methods in conjunction with one another is currently recommended as the preferred way to determine the number of factors underlying the items in a scale (Fabrigar *et al.*, 1999; Hayton *et al.*, 2004). The scree test involves plotting the eigenvalues produced by the PCA from highest to lowest and

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[1] The factor analysis was also re-run using principal factors with prior estimation of communalities using the squared multiple correlation coefficient. The overall shape of the scree plot and parallel analysis results, along with the factor pattern and loadings for the final 8-factor solution were, as expected, almost identical to those obtained with the PCA. However, it should be noted that the scree test and parallel analysis using the principal factors method produced plots that were somewhat more ambiguous than those obtained using PCA. The results of the principal factors analysis are available upon request.

analyzing the shape of the resulting curve (DeVellis, 2003). The point at which the values become horizontal (i.e., the "elbow") corresponds to the point at which additional factors are no longer capturing a substantial amount of information in the items, and factors that lie above the elbow are subsequently retained. A less subjective approach that also has been found to perform well but has been largely underutilized due to its statistical programming requirements (Fabrigar et al., 1999; Hayton et al., 2004; Kabacoff, 2003) is parallel analysis (PA). Parallel analysis involves comparing eigenvalues from study data to eigenvalues obtained from randomly generated data. Multiple random datasets with the same characteristics (i.e., sample size and number of variables) of the study dataset are created and the exploratory factor analysis is run on each of these random datasets. The eigenvalues produced by these random datasets are then computed and compared to the actual eigenvalues produced by the study data. The number of factors to retain is equal to the number of study eigenvalues that exceed the randomly-produced eigenvalues. This approach is thought to be fairly accurate in determining the number of meaningful common factors because it takes into account the effect of sampling error on eigenvalues (Hayton et al., 2004). In this study, we used SAS programming code recently published for conducting parallel analysis (Kabacoff, 2003) to compare the sample eigenvalues to the 95th percentile eigenvalues produced from 100 randomly generated datasets. The results of the scree test and parallel analysis were also evaluated in terms of their parsimony and plausibility to help ensure that the resulting factor model made sense conceptually.

   After determining the number of factors to extract, an oblique (promax) rotation was performed to determine which items best represent, or load most highly on, which common factor. An oblique rotation, which allows factors to correlate with one another, was deemed

appropriate in light of previous research suggesting that domains of self-care and adherence are often modestly but significantly correlated with one another (Hays, 1993; Johnson, 1992; Toobert & Glasgow, 1994). After rotation, items were retained if they achieved factor loadings greater than .40 on their primary factor and no loading greater than .20 on any secondary factor.

   Reliability assessment. Item sets were then assessed for internal consistency reliability by computing Cronbach's alpha, which partitions the variance of the item composite into communal and unique variance. Alpha quantifies the proportion of communal variance in the item set, and values higher than .70 are generally desired. Next, items with poor item-total correlations were eliminated from their subscale in an attempt to optimize scale brevity while maintaining acceptable internal consistency reliability. Finally, an unweighted total score was computed for each subscale by computing the mean of all retained items within the subscale.

   The test-retest reliability of the adherence measure was then computed using data from the 45 respondents who completed the measure a second time, several weeks after the first administration. The Pearson correlation between individuals' scores from the first administration and their scores from the second administration was computed for each subscale to determine the temporal stability of the measure.

   Construct validity assessment. We also assessed evidence for construct validity of the VSMS. One way of assessing construct validity is to evaluate the relationship of scores on the measure in question (i.e., the VSMS) to scores on established measures of other constructs (e.g., current disease activity, depressive symptoms, social support) theorized to be related to the construct in question (i.e., self-management behavior; DeVellis, 2003). Two

types of construct validity, convergent and discriminant validity, were assessed in this study by calculating Pearson correlations between patients' scores on the VSMS subscales and the validation measures described above, and comparing these values to the hypothesized pattern of correlations shown in Table 4.1. Statistically significant positive or negative correlations of the VSMS subscales with measures of other constructs (e.g., self-efficacy, social support, depressive symptoms) empirically related to adherence in previous research (WHO, 2003; Hays 1993) were considered evidence of convergent validity. Null correlations of the VSMS subscales with measures of other constructs (e.g., social desirability, age, gender) empirically *not* related to adherence in previous research (WHO, 2003) were considered evidence of discriminant validity.

Results

Sample Characteristics

Descriptive statistics for the study sample, including socio-demographics, clinical variables, and other important study variables, are shown in Table 4.2. Just over half of the sample (53.7%) was female, and the vast majority (93.6%) reported being White/Caucasian. The mean age of the sample was 54.7 years (*SD*=14.7, range 18-83), and the mean level of education was 14.6 years (*SD*=2.4), or some college. The majority of the sample (70.7%) reported a diagnosis of Wegener's granulomatosis, and the mean duration of living with ANCA-SVV was 75.6 months (*SD*=70.7), or just over six years (range=3 months to 41.6 years). The average score on the General Health scale of the SF-36 was 45.0 (*SD*=23.6), which is substantially lower than the mean of 71.9 reported for the general U.S. population (J.E. Ware, Jr., 2000), but similar to that reported for patients with rheumatoid arthritis (*M*=44, *SD*=23; Ruta, *et al*., 1998). Patients rated their current disease activity significantly

higher than their physicians (t=4.33, p<.0001), but patient and physician ratings were moderately correlated (r=.39, p<.0001). Patients were taking an average of 6.8 prescribed medications for reasons related to ANCA-SVV and had been recommended, on average, about 6 of the 8 possible adherence behaviors assessed in this study. Finally, the average score on the CES-D was 10.7 (SD=9.1), which represents a significantly higher level of depression than that reported for a general community sample (Radloff, 1975, M=9.13, SD=8.27, n=1672; t=2.53, p<.05) (Radloff, 1975). Twenty-five percent of the ANCA-SVV patients in this sample scored above the cutoff of 16 on the CES-D (Radloff, 1977) for depressive symptoms (not shown in table), which is similar to the prevalence for increased depressive symptoms in individuals with primary systemic vasculitis previously reported (Koutantji *et al.*, 2003).

We tested whether the sub-sample of 44 patients who completed the Time 2 questionnaire differed significantly from the rest of the sample on any of the characteristics shown in Table 4.2, using bivariable linear regression. No significant differences were revealed, with one exception. Patients who completed the Time 2 questionnaire rated their current disease activity lower than patients who only completed the Time 1 questionnaire (unstandardized regression coefficient [b]=-.99, standard error [se]=.35, p<.01). That is, Time 2 participants rated their current disease activity, on average, almost one point lower (M=2.16, SD=1.5) than participants who only completed the Time 1 questionnaire (M=3.15, SD=2.2). However, it should be noted that this association was not seen for physician ratings of disease activity or scores on the General Health Scale.

Factor Structure

The eigenvalues for the study sample and the 100 simulated datasets are plotted in Figure 1. The results of the scree test revealed two elbows: one suggesting a nine-factor solution and one suggesting a seven-factor solution. The parallel analysis suggested a nine-factor solution, although the eighth and ninth eigenvalues for the study sample were just slightly higher than the eighth and ninth eigenvalues for the simulated datasets. Because of the ambiguity of these results, we examined solutions for seven, eight, and nine factors.

The eight-factor solution resulted in the fewest number of cross-loadings and distinct self-management factors that made sense theoretically, and accounted for 58.2% of the total variance (compared to 55.1% for the seven-factor solution and 61.2% for the nine-factor solution). The eight factors identified by this solution included six adherence domains (adherence to recommended health services, exercise adherence, symptom monitoring adherence, medication adherence, infection avoidance adherence, and diet adherence), as well as two general self-care domains (prompt reporting of symptoms and side effects to a health professional and adjusting activity level in response to symptoms). The rotated factor loadings for all items are shown in Table 4.3. The seven-factor solution differed from the pattern shown in Table 4.3 in that items pertaining to limiting activities and diet adherence tended to load on one factor, with the diet items then cross-loading on the exercise factor as well. The nine-factor solution resulted in two factors for use of health services: one for attending appointments with physicians and one for obtaining tests and immunizations. However, items within these two factors tended to cross-load on both factors, and additional cross-loadings were then seen for items loading primarily on the other seven factors. Thus, the eight-factor solution was accepted as the final solution, and 14 items with cross-loadings higher than .20 on any other factor were then deleted from the eight subscales.

The inter-factor correlations are shown in Table 4.4. The highest observed correlations were found between exercise adherence and symptom monitoring adherence (r=.32) and for medication adherence and adherence to recommended health services (r=.31).

Examination of the item-total correlations and the impact on Cronbach's $\acute{\alpha}$ of deleting each item led us to delete one item from the exercise adherence scale that resulted in an improvement in alpha. We also dropped three of the 10 remaining items from the reporting symptoms and side effects scale to decrease the length of this scale while maintaining $\acute{\alpha}$ >.90. Correlations between the final 8 subscales are shown in Table 4.5. The general pattern of inter-factor correlations is similar to those shown in Table 4.4, although correlations between the final subscales tended to be slightly larger than correlations between the raw subscales in the factor analysis.

Test-Retest Reliability

Descriptive statistics (*n*'s, percent missing, *M*, *SD*, and $\acute{\alpha}$) for the final eight subscales for both Time 1 and Time 2 are shown in Table 4.6, along with the correlation between subscale scores at Time 1 and Time 2. The test-retest reliability for all eight subscales ranged from .56 (*p*<.001) for adherence to recommended health services to .79 (*p*<.001) for exercise adherence.

Construct Validity

The Pearson correlations for the eight self-management subscales with the key variables for assessing construct validity are shown in Table 4.7.

Expected null correlations: Discriminant validity

Correlations between the eight self-management subscales and age, gender, and social desirability were expected to be non-significant. As expected, age was not significantly

correlated with six of the eight subscales, but unexpectedly, was significantly and positively

associated with two subscales: medication adherence (r=.33, p<.001) and adjusting activities

in response to fatigue and symptoms (r=.30, p<.001). Also as expected, gender was not

associated with seven of the eight subscales. However, gender was significantly associated

with medication adherence. Contrary to our hypothesis, males demonstrated significantly

higher medication adherence than females (r=.30, p<.001). Finally, contrary to our

hypotheses, scores on five of the eight subscales were modestly but significantly associated

with scores on the social desirability scale, with correlations ranging from r=.15 (p<.05) for

adherence to recommended health services to r=.26 (p<.001) for medication adherence.

Only exercise adherence, infection avoidance adherence, and diet adherence demonstrated

the expected non-significant associations with social desirability.

Expected significant correlations: Convergent validity

Observed correlations between the socio-demographic, clinical, and regimen

characteristics and the eight self-management subscales generally did not conform to

predictions, with a few exceptions. Contrary to hypotheses, education was not significantly

associated with six of the eight subscales and was actually negatively associated with two of

the subscales: adherence to recommended health services (r=-.14, p<.05) and medication

adherence (r=-.21, p<.01). Also contrary to hypotheses, none of the three measures of

disease activity or damage were significantly correlated with any of the eight subscales. The

modest and negative correlations expected between the three indicators of treatment

complexity and the self-management subscales were also not observed, with the exception of

one: the correlation between patient-perceived treatment complexity and medication

adherence (r=-.27, p<.001). Finally, disease duration was negatively correlated with the

medication adherence (r=-.14, p<.05) and adherence to recommended health services factors (r=-.19, p<.05), but unexpectedly, was not significantly correlated with the other six subscales. This pattern of findings did not change substantively after controlling for socio-demographics (age, gender, and education) and social desirability.

Observed correlations between the psychosocial and behavioral variables and the eight self-management subscales conformed more closely to the expected pattern, with some mixed results. General adherence showed a more expected pattern of associations, in that it was positively and significantly correlated with five of the eight subscales: reporting symptoms and side effects (r=.20, p<.01), exercise adherence (r=.39, p<.001), symptom monitoring adherence (r=.26 p<.01), medication adherence (r=.26, p<.001), and diet adherence (r=.31, p<.001). These relationships persisted even after controlling for social desirability, which because of its positive relationships to both general adherence and several of the subscales of the VSMS, had the potential to act as a confounding variable. Social support was modestly and positively correlated with four of the eight subscales (reporting symptoms and side effects, adherence to recommended health services, symptom monitoring adherence, and diet adherence), but was not significantly related to the other four subscales. Self-efficacy was not associated with six of the eight subscales but was correlated with two subscales in the expected direction: medication adherence (r=.16, p<.05) and diet adherence (r=.16, p<.05). As expected, depression was negatively and significantly correlated with three of the eight subscales (adherence to recommended health services, medication adherence, and diet adherence) but was not significantly associated with any of the other five self-management subscales.

Discussion

To our knowledge, this is the first study of illness self-management in adult patients living with ANCA-SVV. We aimed to develop a self-report measure that conveniently assesses the self-care behaviors relevant for the majority of adult patients living with ANCA-SVV. We also aimed to gather preliminary evidence of the measure's reliability and construct validity.

As expected, multiple domains of self-care behavior were identified as relevant to these patients' illness self-management, and the VSMS reflected the multi-dimensionality of this construct. Although initially we assessed 11 different types of self-care behavior, including 8 adherence behaviors and 3 general self-care behaviors, our analysis suggested eight behavioral domains that are relevant for the majority of ANCA-SVV patients. These eight domains included six adherence behaviors recommended by health professionals for a majority of patients (medication adherence, use of recommended health services, infection avoidance adherence, diet adherence, exercise adherence, and symptom monitoring adherence), and two general self-care behaviors relevant for all ANCA-SVV patients (prompt reporting of symptoms and side effects, and adjusting activity levels in response to symptoms). One hypothesized domain, adherence to medications administered by health professionals, was found to pertain to only a minority (approximately 25%) of patients with ANCA-SVV. The lack of applicability of this behavior to the illness situations of the majority of patients in our study may be related to the fact that our sample, on average, had been living with ANCA-SVV for several years and as a result no longer required the intravenous medication often used to attain an initial remission. As a result, items assessing this behavior were dropped from subsequent analyses and the final self-management measure. Future studies may want to evaluate these items with a larger sample of ANCA-SVV patients

currently taking medication administered by a health professional. In addition, although we initially thought that items assessing patients' use of health services might fall into two domains, one for appointment-keeping with health professionals and one for obtaining tests and immunizations, the pattern of factor loadings suggested one factor capturing both behaviors. However, it is important to note that the items assessing appointment-keeping had loadings for the health services factor that were somewhat lower than the items assessing medical test and immunization behavior. In addition, all but one of the items asking about appointment-keeping exhibited small but relevant cross-loadings on other factors and were therefore dropped from the final scale. In light of the cross-loadings for these items, along with the somewhat ambiguous results of the scree test and parallel analysis, researchers using this scale in the future may want to retain the discarded appointment-keeping items and re-examine the factor structure to confirm that these items again load on one factor along with items for obtaining tests and immunizations. Finally, while we initially allowed items assessing patients' prompt reporting of symptoms to load separately from items assessing prompt reporting of medication side effects, these items, not surprisingly, loaded together on one factor.

As expected, correlations between behavioral domains ranged from not significantly different than zero, to positive and moderate in magnitude. None of the correlations, either those from the factor analysis or those only using the final subscales, were greater than $r=.37$ ($p<.001$), which is well below the cutoff of $r=.70$ considered to be high enough to result in factor interpretation errors (Nunnally, 1978). In addition, the magnitude and direction of significant inter-factor correlations are consistent with prior research on treatment adherence and self-care behavior (WHO, 2003). For example, Toobert and colleagues (Toobert &

Glasgow, 1994) found correlations between diet and exercise adherence of r=.27 to r=.34, comparable to the correlation of r=.35 found between the final diet and exercise adherence subscales in this study.

Also as expected, examination of Cronbach's $\acute{a}$ revealed that internal consistency reliability ranged from adequate to excellent for seven of the eight VSMS subscales. Three scales demonstrated excellent internal consistency reliability, with alphas greater than .90: reporting symptoms and side effects, exercise adherence, and symptom monitoring adherence. The subscale with minimally acceptable internal consistency reliability, defined as between .65 to .70 (DeVellis, 2003), was the subscale for adjusting activities in response to symptoms (Time 1 $\acute{a}$=.67). However, the alpha for this scale increased to above .70 at Time 2, and test-retest reliability was high (r=.77, p<.001). In general, the temporal stability of all eight subscales was also good, with test-rest correlations ranging from .56 for adherence to recommended health services to .79 for exercise adherence. It is widely acknowledged that test-retest reliability estimates are imperfect for assessing the reliability of a measure for several reasons; e.g., test-retest correlations may be reduced if real changes in individuals' self-management behavior occur across the interval between administrations, or they may be increased due to memory effects if the interval between administration is too short. In this study, the average of 6.2 weeks between administration makes the former possibility more likely. Even in light of this, the test-retest correlations were large in magnitude. Taken together, the alphas and test-retest correlations suggest that the self-management measure has good reliability.

The pattern of correlations between the VSMS subscales and socio-demographic, clinical, regimen, psychosocial, and behavioral characteristics demonstrated mixed support for the

construct validity of the measure. Convergent validity was supported by the direction of the observed correlations of many of the VSMS subscales (in particular, the medication adherence, diet adherence, symptom monitoring adherence, use of recommended health services, and reporting symptoms and side effects subscales) with the psychosocial and behavioral variables (e.g., general adherence, social support, self-efficacy, and depressive symptoms), although not all correlations were significant as expected. The non-significant correlations of age and gender with most of the VSMS subscales provides some evidence of discriminant validity, although unexpected significant correlations were found for medication adherence and male gender, as well as for age and three of the subscales (medication adherence, diet adherence, and adjusting activities in response to symptoms). Finally, it is particularly promising that scores on the General Adherence Scale, a measure with good evidence of construct and predictive validity when used with Type II diabetes, hypertension, and heart disease patients (Kravitz et al., 1993) as well as cancer patients (DiMatteo *et al.*, 1993), correlated positively with all eight subscales, and was moderately and significantly correlated with five subscales, even after controlling for social desirability. That is, the relationship between general adherence and the five VSMS subscales could not be explained by patients' tendency to report their behavior in socially desirable ways. These significant observed relationships are further evidence of convergent validity for the VSMS.

It is somewhat concerning that, for the most part, we did not observe the expected relationships between the VSMS subscales and the clinical (e.g., ratings of disease activity and damage) and regimen characteristics (e.g., ratings of treatment complexity, number of medications, and number of behavioral recommendations). However, most of these variables were measured using one-item measures created specifically for this study that have

unknown psychometric properties (10-point ratings of disease activity and damage, indicators of treatment complexity). Using measures for these clinical and regimen characteristics with unknown reliability or validity may have compromised our ability to see the modest relationships expected between them and the VSMS. It should be noted that the more established measures used in this study (e.g., General Adherence Scale, Social Provisions Scale, self-efficacy, and the CES-D) tended to conform to the expected pattern of correlations more so than these one-item measures. In addition, the medication adherence subscale was significantly associated, in the expected direction and magnitude, with disease duration and perceived treatment complexity, providing further evidence of validity for this particular subscale.

Unexpected results were also found for the relationship between education and scores on the self-management subscales, which were either null or in the opposite direction of our hypotheses, as well as for scores on the social desirability scale with scores on five of the eight self-management subscales. However, these latter correlations tended to be small (under $r=.20$ for three of the scales, with the highest correlation being $r=.26$ for medication adherence), and similar to those found with other self-report measures of self-care behavior with otherwise good evidence of validity (Hays, 1993). Future users of the VSMS may want to also assess social desirability and control for this possible source of bias in their analyses.

The mixed evidence for construct validity points to the need for more research using this measure. In particular, comparing scores on the VSMS subscales to objective measures of adherence (e.g., medication levels in blood, clinic records documenting patients' use of health services) and self-management behavior would further help evaluate the measure's predictive validity. Examining correlations of the scores on the VSMS subscales to objective

measures of disease control (blood and protein levels in urine, ANCA titers, or kidney function) or complications (e.g., incidence of infections) would also help evaluate the measure's validity.

Several limitations to this study should be noted. First, the sample for this study consisted of a convenience sample that was relatively small, especially when missing data was taken into account. Ideally, we would have liked to have at least 200 non-missing data points for all items, with 300 being even better, but the difficulty in recruiting this rare patient population limited our sample size. However, using the pairwise correlation matrix in the factor analysis allowed us to take advantage of all available data, and a plausible factor solution did emerge, despite the small sample. Second, we relied on patient recall for assessing which self-care behaviors had been explicitly recommended to them by a health professional. Thus, the six adherence subscales are actually measuring *perceived* adherence. Future research should compare physician reports of behavioral recommendations to patient recall of recommendations reported using this measure, to determine rates of discrepancies, and in turn, unintentional non-adherence by patients due to a failure to recall or understand recommendations. Third, the lack of prior research on illness self-management in ANCA-SVV and some of the self-care behaviors assessed in the new self-management measure makes it difficult to determine if the mixed pattern of support for construct validity is due to an actual lack of construct validity or real differences in relationships of these variables to the different types of self-care behaviors involved in ANCA-SVV self-management. It may be that influences on self-care behavior are different for this patient population, or vary for different types of behavior. Given that the majority of research on treatment adherence has focused on medication adherence, to the exclusion of other types of self-care behavior (e.g.,

communicating symptoms promptly to a health professional, taking steps to avoid infection),

and that different types of self-care behavior are relatively independent of one another, it is

plausible to imagine that the correlates of self-care behavior may vary, to some degree,

across specific behaviors.

Despite these limitations, this study presents a promising approach for measuring illness

self-management in patients living with ANCA-SVV, an extremely under-studied patient

population. The measure developed in this study is a convenient way to assess a

comprehensive set of self-care behaviors relevant for most patients trying to manage ANCA-

SVV, and is also flexible for use in a variety of patients who may be asked to follow a range

of treatment recommendations. Furthermore, the self-report nature of the VSMS allows for

easy assessment of self-care behaviors not easily measured using objective measures (e.g.,

infection avoidance behavior, exercise adherence, diet adherence). The multi-dimensionality

of the measure conforms with what is known about self-care behavior in other patient

populations, and reliability of the eight self-management subscales appears good. While

preliminary evidence for the construct validity of the measure is mixed and by no means

complete, future research can build on the findings of this study to more thoroughly evaluate

validity. If this evidence further supports the reliability and validity of the measure, it will

likely prove useful in both clinical and research settings. For example, its use in clinical

settings has the potential to help physicians treating ANCA-SVV patients identify and

resolve problems with self-care (Rand, 2000). In addition, its use in research settings has the

potential to identify important influences on self-care in this illness context and inform the

development of interventions for this patient population.

Table 4.1. Direction and magnitude of hypothesized relationships between validation
measures and vasculitis-specific self-management behavior.

Validation Measures	Predicted direction of association	Predicted magnitude of association
Socio-demographics		
Age	Zero	Zero
Gender	Zero	Zero
Education	Positive	Small
Clinical and health variables		
Duration of diagnosis	Negative	Small to moderate
Current disease activity	Positive	Small to moderate
Disease damage	Positive	Small to moderate
Regimen characteristics		
Perceived treatment complexity	Negative	Small to moderate
Number of prescribed medications	Negative	Small to moderate
Number of recommendations	Negative	Small to moderate
Psychosocial and behavioral characteristics		
Social desirability	Zero	Zero
General adherence to medical recommendations	Positive	Small
Perceptions of available social support	Positive	Small to moderate
Vasculitis-specific self-efficacy	Positive	Small to moderate
Depressive symptoms	Negative	Moderate

Table 4.2. Descriptive statistics for ANCA-SVV patients.

	% or M (SD)	
	Full sample (*n*=205)	Time 2 sample (*n*=44)
Socio-demographics		
Gender		
Male	46.3%	40.9%
Female	53.7%	59.1%
Race/ethnicity		
White	93.6%	93.0%
American Indian/Alaskan Native	1.0%	0%
Asian	2.5%	2.3%
Black or African American	1.0%	2.3%
Hispanic/Latino	2.0%	2.3%
Age, in years	54.7 (14.7)	56.5 (14.8)
Years of education	14.6 (2.4)	15.1 (2.3)
Marital status		
Married	74.2%	75.0%
Non-married	25.9%	25.0%
Clinical characteristics		
Diagnosed condition		
Wegener's granulomatosis	70.7%	68.2%
Microscopic polyangiitis	7.8%	9.1%
Churg Strauss syndrome	4.9%	4.6%
ANCA-glomerulonephritis	15.1%	18.2%

Good Pasture's disease	1.0%	0%
Temporal arteritis	0.5%	0%
Time since diagnosis, in months	75.7 (70.7)	88.5 (88.3)
Time between symptom onset and diagnosis, in months	13.9 (34.0)	12.7 (25.8)
Patient-perceived disease activity, range 1-10	2.9 (2.1)	2.2 (1.5)
Physician-perceived disease activity, range 1-10	1.9 (1.3)	1.9 (1.2)
Physician-perceived damage, range 1-10	4.4 (2.6)	4.6 (2.6)
General physical health, range 0-100	45.0 (23.6)	50.6 (22.7)
Ever on dialysis	15.8%	16.3%
Currently on dialysis	3.5%	0%
History of kidney transplant	7.5%	11.4%
Regimen characteristics		
Patient-perceived regimen complexity, range 1-10	2.9 (2.5)	2.4 (2.0)
Number of prescribed medications	6.8 (4.1)	6.5 (3.9)
Number of recommended behaviors, range 0-8	5.9 (1.4)	6.0 (1.3)
Psychosocial characteristics		
General adherence, range 1-6	5.2 (0.7)	5.3 (0.7)
Social support, range 24-144	128.3 (14.4)	130.6 (11.9)
Self-efficacy, range 10-100	67.7 (16.7)	71.9 (15.1)
Depressive symptoms, range 0-60	10.7 (9.1)	29.2 (6.8)
Social desirability, range 1-20	12.5 (3.8)	12.1 (3.7)

Table 4.3. Promax rotated factor structure for the 62-item Vasculitis Self-Management Scale: 8 factor solution.

	Factor 1: Reporting symptoms & side effects	Factor 2: Health services adherence	Factor 3: Exercise adherence	Factor 4: Symptom monitoring adherence	Factor 5: Medication adherence	Factor 6: Infection avoidance adherence	Factor 7: Diet adherence	Factor 8: Adjusting activity
Med1					.70			
Med2					.57			
Med3					.62			
Med4					.83			
Med5					.80			
Med6†			-.24		.83			
Med7					.53			
Doctor1†		.48			.37	-.21		
Doctor2†		.50			.30		.21	
Doctor3†		.42			.29		.24	
Doctor4		.54						
Doctor5†		.49						.26

Doctor6†		.39	.26	.22
Test1		.80		
Test2		.83		
Test3		.71		
Test4†		.64		-.24
Test5†		.78	-.23	.23
Test6		.67		
Act1				.55
Act2				.65
Act3				.64
Act4†			.23	.63
Act5				.70
Act6				.42
Sym1	.81			
Sym2	.68			

	Col1	Col2	Col3	Col4	Col5
Sym3	.80				
Sym4†	.71				.22
Sym5*	.58				
Sym6	.70				
Side1	.83				
Side2	.73				
Side3	.80				
Side4*	.71				
Side5*	.59				
Side6†	.67			.21	-.22
Infect1				.79	
Infect2				.82	
Infect3†			.21	.62	
Infect4†		.25	-.36	.41	
Infect5				.65	
Infect6				.81	

Infect7					.76	
Diet1						.65
Diet2						.72
Diet3†	-.22					.69
Diet4						.65
Diet5						.66
Diet6†				-.23		.67
Exer1		.87				
Exer2		.91				
Exer3		.87				
Exer4		.86				
Exer5		.82				
Exer6*		.68				
Mon1			.76			
Mon2			.84			

88

Mon3	.88
Mon4	.79
Mon5	.82
Mon6	.81

†Items dropped due to cross-loadings on a secondary factor; *Items dropped after examination of item-total correlations and alpha.

Table 4.4. Inter-factor correlations for 8-factor solution.

	Factor 1: Reporting symptoms and side effects	Factor 2: Health services adherence	Factor 3: Exercise adherence	Factor 4: Symptom monitoring adherence	Factor 5: Medication adherence	Factor 6: Infection avoidance adherence	Factor 7: Diet adherence	Factor 8: Adjusting activity
Factor 1: Reporting symptoms and side effects	1.0	--	--	--	--	--	--	--
Factor 2: Health services adherence	.26	1.0	--	--	--	--	--	--
Factor 3: Exercise adherence	.06	.01	1.0	--	--	--	--	--
Factor 4: Symptom monitoring adherence	.28	.09	.32	1.0	--	--	--	--
Factor 5: Medication adherence	.26	.31	.09	.17	1.0	--	--	--
Factor 6: Infection avoidance adherence	.20	-.07	-.003	.13	.08	1.0	--	--
Factor 7: Diet adherence	.11	.17	.23	.23	.24	.14	1.0	--
Factor 8: Adjusting activity	.16	.06	.03	.14	.14	.19	.21	1.0

Table 4.5. Pearson correlations between the 8 final self-management subscales.

	Factor 1: Reporting symptoms/side effects	Factor 2: Health services adherence	Factor 3: Exercise adherence	Factor 4: Symptom monitoring adherence	Factor 5: Medication adherence	Factor 6: Infection avoidance adherence	Factor 7: Diet adherence	Factor 8: Adjusting activity
Factor 1: Reporting symptoms/side effects	1.0	--	--	--	--	--	--	--
Factor 2: Health services adherence	.26***	1.0	--	--	--	--	--	--
Factor 3: Exercise adherence	.07	.06	1.0	--	--	--	--	--
Factor 4: Symptom monitoring adherence	.34***	.08	.37***	1.0	--	--	--	--
Factor 5: Medication adherence	.25**	.23**	.21*	.19*	1.0	--	--	--
Factor 6: Infection avoidance adherence	.20*	-.04	-.03	.09	.17	1.0	--	--
Factor 7: Diet adherence	.12	.12	.35***	.21*	.19*	.24**	1.0	--

| Factor 8: Adjusting activity | .19* | -.003 | .07 | .16 | .15* | .25** | .33*** | 1.0 |

*p<.05
**p<.01
***p<.001

Table 4.6. Means, standard deviations, internal consistency reliability, and test-retest reliability for 8 subscales, at Times 1 and 2.

	Time 1				Time 2				
	N	% missing	Mean (SD)	Alpha	N	% missing	Mean (SD)	Alpha	Test-retest correlation
Factor 1: Reporting symptoms/side effects	177	14%	3.2 (1.08)	.90	40	11%	3.1 (1.02)	.86	.76*
Factor 2: Health services adherence	202	1%	4.6 (.63)	.78	44	2%	4.6 (.58)	.73	.56*
Factor 3: Exercise adherence	125	39%	3.2 (1.02)	.94	29	36%	2.8 (.94)	.96	.79*
Factor 4: Symptom monitoring adherence	133	35%	3.8 (.97)	.91	29	36%	3.5 (1.07)	.94	.68*
Factor 5: Medication adherence	191	7%	4.5 (.54)	.77	43	4%	4.6 (.52)	.87	.60*
Factor 6: Infection avoidance adherence	142	31%	3.7 (.79)	.85	33	27%	3.9 (.73)	.84	.67*
Factor 7: Diet adherence	154	25%	3.6 (.76)	.76	34	24%	3.6 (.73)	.71	.65*
Factor 8: Adjusting activity	185	10%	3.4 (.64)	.67	40	11%	3.5 (.62)	.71	.77*

*p<.001

Table 4.7. Pearson correlations with construct validity variables[†].

	Factor 1: Reporting symptoms and side effects	Factor 2: Health services adherence	Factor 3: Exercise adherence	Factor 4: Symptom monitoring adherence	Factor 5: Medication adherence	Factor 6; Infection avoidance adherence	Factor 7: Diet adherence	Factor 8: Adjusting activity
Socio-demographics								
Age (0)	.03	.09	.06	.03	.33***	.08	.21*	.30***
Gender (0)	.11	-.05	.10	.13	.30***	.06	.11	.02
Education (+)	-.11	-.14*	.04	-.14	-.21**	.06	.08	-.07
Clinical and health variables								
Duration of diagnosis (-)	-.10	-.19*	.09	.03	-.14*	-.07	.05	.03
Physician perceived disease activity (+)	.05	.12	.20	.15	-.01	-/07	.08	-.05
Patient perceived disease activity (+)	-.12	.04	-.09	-.07	-.08	-.01	-.11	-.06
Physician perceived damage (+)	.03	.03	.11	.03	.03	.08	-.11	.05

Regimen characteristics

Perceived treatment complexity (-)	.0008	-.02	-.06	.004	-.27***	.14	.08	.03
Number of medications (-)	-.04	.03	-.04	.08	-.09	.16	.07	.06
Number of recommendations (-)	.04	.12	-.06	.15	-.06	.14	.09	.12

Psychosocial and behavioral characteristics

Social desirability (0)	.19*	.15*	.05	.22*	.26***	.15	.14	.17*
General adherence (+)	.20**	.13	.39***	.26**	.26***	.10	.31***	.05
Social support (+)	.16*	.18**	.04	.19*	.14	.06	.20*	.08
Self-efficacy (+)	.13	.05	.03	.03	.16*	.05	.16*	.007
Depressive symptoms (-)	-.09	-.14*	.01	-.11	-.19**	-.004	-.18*	.007

†Expected direction of correlation for each variable is shown in parentheses in Column 1.
*p<.05
**p<.01
***p<.001

Figure 4.1. Eigenvalues for real sample data and 100 simulated datasets.
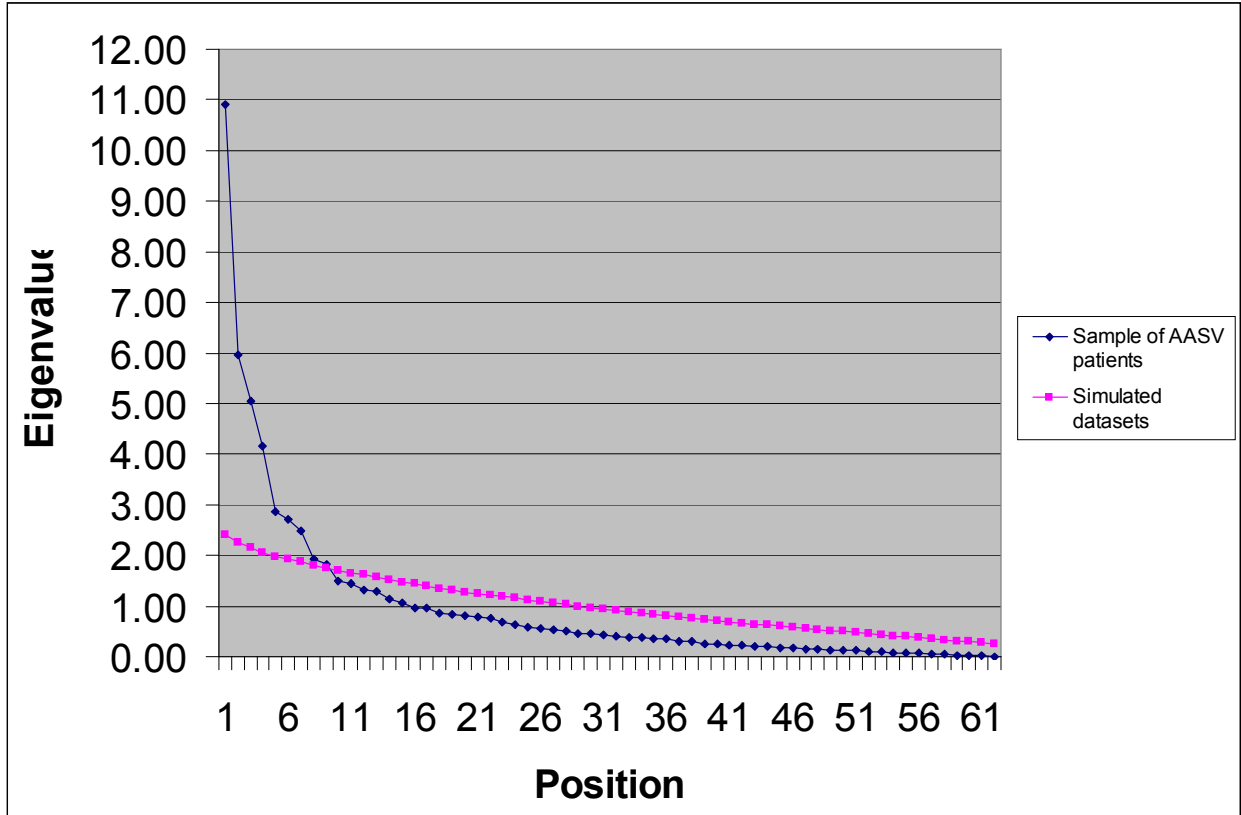
CHAPTER FIVE: AN EXPLORATORY ANALYSIS OF PERCEIVED BARRIERS AND FACILITATORS TO ILLNESS SELF-MANAGEMENT AMONG ADULTS LIVING WITH ANCA-ASSOCIATED SMALL VESSEL VASCULITIS

Abstract

Objective: To characterize attitudes and beliefs, including perceived barriers and facilitators, about performing the self-management behaviors among adults living with ANCA-associated small vessel vasculitis (ANCA-SVV), and determine if these attitudes and beliefs were associated with levels of self-management behavior.

Methods: A total of 205 adults living with ANCA-SVV or a closely related condition completed a self-administered questionnaire that included measures of self-management behavior, perceptions of barriers and facilitators to performing recommended behaviors, social desirability bias, and a variety of socio-demographic, clinical, and health variables. Descriptive statistics were generated to describe, for each of 11 self-management behaviors, perceived difficulty, total number of perceived barriers, perceived importance, total number of facilitators, and specific perceived barriers and facilitators reported by at least 10% of patients. Regression analyses explored whether these variables were associated with patients' self-reported performance of each self-management behavior, controlling for several potential confounding variables.

Results: With a few exceptions, perceived difficulty and total number of perceived barriers were consistently and negatively associated with self-management behavior, while greater perceived importance predicted higher levels of self-management. Patients' reporting of a number of specific barriers was negatively associated with performance of each self-management behavior, but reporting of specific facilitators was not significantly related to greater performance of self-management behavior.

Conclusions: This study reveals that as with other patient populations, patient attitudes and beliefs about their illness and its treatment influence self-management behavior in adults

living with ANCA-SVV. Perceived barriers and facilitators to adherence to medication, recommended health services, diet, and exercise were similar to those found in other patient populations. This study also provides new insights into barriers and facilitators experienced by patients in performing additional self-management behaviors (e.g., infection avoidance, reporting symptoms) not often studied in prior research. Implications for further research and intervention development are discussed.

Introduction

ANCA-associated small vessel vasculitis (ANCA-SVV) is a group of rare autoimmune conditions, characterized by inflammation and necrosis of blood vessels primarily in the respiratory tract and kidneys (Watts & Scott, 1997), with an overall incidence estimated around 39 per million (Watts *et al.*, 1995). ANCA-SVV is a potentially fatal, relapsing illness, with an often progressive but unpredictable course (Koutantji *et al.*, 2000). Until recently, rapid death from ANCA-SVV was likely (Fauci *et al.*, 1983), but the introduction of effective immunosuppressive medications has transformed ANCA-SVV into a chronic illness with which most patients live for many years (Koutantji et al., 2000). There is no cure for the illness, but treatment with immunosuppressive medications induces remission in 80-100% of patients (Langford, 2003). Patients may experience full remission, partial remission, and/or minor or major relapses (Jayne, 2003). Even among patients in remission, the risk of relapse and morbidity from the disease and its ongoing, highly toxic treatment remains high (Hoffman *et al.*, 1998).

As with other serious chronic illnesses, individuals who are diagnosed with ANCA-SVV face a daunting array of psychosocial challenges. In addition to living with an unpredictable and potentially fatal prognosis, patients and their families are asked to undertake complicated treatment regimens that come with considerable risk of serious side effects (Jayne, 2003) and require substantial role adjustment and lifestyle changes (Koutantji et al., 2000). Yet leading researchers have noted the paucity of research on the psychosocial aspects of living with vasculitis. In an editorial outlining some of the unanswered questions about the psychological aspects of vasculitis, Koutantji, Pearce, and Harrold (Koutantji et al., 2000) suggested that a major goal of future research should be to evaluate coping, functioning, and

quality of life issues in patients living with vasculitis, all issues that have been under-studied in this patient population.

One of these issues not previously studied is patients' illness self-management, defined as the "day-to-day tasks an individual must undertake to control or reduce the impact of disease on physical health status" (Clark *et al.*, 1991). Patients assume these tasks with the collaboration and guidance of their health care providers (Clark et al., 1991). A construct related to illness self-management is treatment adherence, or "the extent to which a person's behavior – taking medication, following a diet, and/or executing lifestyle changes – corresponds with agreed recommendations from a health care provider" (WHO, 2003). Although many self-management tasks include adhering to physicians' recommendations, self-management is a broader construct of which treatment adherence is one aspect (Gallant, 2003). Self-management involves mastery of three types of activities: performing activities to manage the condition, making informed decisions about care, and applying skills to maintain adequate psychosocial functioning (Clark et al., 1991). Specific self-management behaviors required by patients vary across conditions (Clark et al., 1991).

Self-management of ANCA-SVV has very rarely been researched. This gap in the literature is concerning, given that poor adherence in this population can lead to renal failure (Andrassy *et al.*, 1991) and presumably, further illness complications, impaired quality of life, and even death. To our knowledge, the Vasculitis Self-Management Study, in which a self-report measure of self-management behavior designed for adults living with ANCA-SVV was developed and tested (Thorpe, et al., in preparation), is the first systematic investigation of self-management among ANCA-SVV patients. In this study, the researchers identified and measured, using the Vasculitis Self-Management Scale (VSMS), eight domains of self-

management behavior that are relevant for the majority of ANCA-SVV patients: 1) self-administered medication adherence; 2) adherence to recommended health services, including appointments with health professionals, medical tests, and immunizations; 3) adherence to infection avoidance recommendations; 4) dietary adherence; 5) exercise adherence; 6) adherence to symptom monitoring recommendations; 7) adjusting activity level in response to fatigue or symptoms; and 8) prompt reporting of new or increased illness symptoms and side effects to a health professional. In addition, some patients, particularly in the initial stage of the illness after diagnosis, are required to take medications administered by a health professional or clinic (e.g., intravenous cyclophosphamide), although a subscale measuring this behavior was not developed in the VSMS. While an important step in the study of self-management among this patient population, this first phase of research stopped short of exploring reasons for non-adherence or sub-optimal performance of ANCA-SVV self-management behaviors, and factors that may influence self-management in this population.

A vast amount of prior research on illness self-management in other patient populations has shown that influences on self-management behavior represent a wide range of factors (WHO, 2003). For example, a recent review on treatment adherence in chronic illness by the World Health Organization (WHO) argued that influences on adherence are multi-dimensional, including factors external to individuals as well as within individuals. In their cross-illness review, they identified a large number of social and economic factors (e.g., poverty, low level of education, lack of social support, rural location), health care team/health system factors (e.g., poor patient-provider relationship, inadequate insurance coverage of costs), condition-related factors (e.g., high level of disability, lack of illness symptoms), therapy-related factors (e.g., complexity of the treatment regimen, medication

side effects), and patient-related factors (e.g., attitudes and beliefs about the illness and treatment, depression) that have been shown to serve as barriers and facilitators to individuals' performance of illness self-management behaviors. The WHO report also found that barriers and facilitators to performing recommended behaviors varied, to some extent, across illnesses (e.g., diabetes versus asthma) and specific behaviors (e.g., medication adherence versus exercise adherence). This is not surprising, given that research has also shown that patients' level of adherence varies across illnesses (Bosworth *et al.*, 2006) and specific recommended behaviors (Bosworth et al., 2006; Johnson, 1992; Toobert & Glasgow, 1994). Furthermore, a recent review on treatment adherence underscored the importance of developing intervention strategies that are tailored to the demands of specific illnesses (Bosworth et al., 2006).

Research and behavioral theory also point to the importance of patients' *perceptions* about the illness and its treatment as determinants of self-management behavior (Bosworth & Voils, 2006; WHO, 2003). For example, the health belief model, the behavioral theory that has been most frequently applied to studying treatment adherence (Bosworth et al., 2006; Leventhal *et al.*, 1984), specifies that patients' perceptions of barriers to performing recommended behaviors directly determine their performance of the behaviors. A variety of other individual perceptions, including beliefs about the severity of the illness or potential outcomes, benefits of the behavior, susceptibility to complications, and confidence in performing the behavior, are also widely believed to affect performance of illness self-management behavior (Bosworth & Voils, 2006).

Thus, the first aim of this study was to characterize patients' attitudes and beliefs about performing the self-management behaviors relevant to ANCA-SVV. Toward this aim, we

developed three specific research questions: 1) Which self-management behaviors do patients generally find most difficult to perform? 2) Which behaviors do they believe are the most important to perform? 3) What are the specific factors that patients perceived as impeding or facilitating the performance of these behaviors? The second aim of this study was to determine if patients' perceptions about self-management behaviors were associated with their self-reported behavior. Specifically, we were interested in learning, for each self-management behavior, whether patients' perceived difficulty and importance, their total number of perceived barriers and facilitators reported, and their reporting of specific barriers and facilitators predicted levels of the self-management behavior.

Methods

Design

This study consisted of a cross-sectional, observational design. All participants completed a self-administered, mailed questionnaire assessing various aspects of ANCA-SVV self-management, psychosocial issues, and socio-demographics. Participants were provided with a $10 gift (in the form of cash or a gift card) after completing and returning the questionnaire.

Sample

To be eligible for the study, patients were required to be 18 years or older, be able to read and write in English, and have received a diagnosis of a form of ANCA-SVV or a closely related condition. Autoimmune conditions that are generally treated using the same types of medication, tend to affect the same organs, and have similar prognoses as ANCA-SVV (e.g., Good Pasture's disease) were included to ease the difficulty of recruiting enough individuals with this rare disease into the study. Adult patients with these conditions were recruited for participation by posting announcements about the study in a variety of places. First, patients

18 years or older who were part of an ANCA-SVV and glomerulonephritis patient disease

registry at the lead author's institution and had signed long-term consent to be contacted

about vasculitis research were invited to participate via letter. Announcements about the

study were also placed in newsletters targeted toward ANCA-SVV patients and on several

ANCA-SVV-related websites. Finally, ANCA-SVV patients who had volunteered for

another study in which the lead author was involved were asked if they were interested in

hearing about other ANCA-SVV research, and if they responded positively, they were

invited to participate in this study. A total of 278 interested patients were identified through

these methods and mailed study consent forms. Of these, 233 (84%) signed and returned the

consent form, and 208 (75%) completed and returned the study questionnaire. Two

participants were subsequently excluded from the analyses because they or their doctor

reported a primary diagnosis of lupus and one additional participant was excluded because he

reported an unknown diagnosis which his physician later indicated was not vasculitis-related.

This resulted in a final sample of 205 patients.

Measures

ANCA-SVV Self-management

ANCA-SVV self-management was measured using the Vasculitis Self-Management Scale

(VSMS). The initial testing of the VSMS was conducted using the current study's sample

and is described in detail, along with pilot work for developing the VSMS, elsewhere

(Thorpe, et al., in preparation). The VSMS was designed to assess individuals' performance

of behaviors directly recommended by health professionals to them for controlling their

condition and maximizing physical and psychosocial functioning, and also their performance

of self-care behaviors generally recommended for all ANCA-SVV patients. Thus, the VSMS

assesses both treatment adherence, traditionally defined as the frequency with which patients'

behavior corresponds with medical recommendations (WHO, 2003), as well as performance

of other relevant self-care behaviors, such as appropriate responding to illness symptoms,

that are important in self-management of ANCA-SVV.

The original version of the VSMS administered to the current patient sample consisted of

68 items that assessed individuals' performance of 11 different behaviors (8 adherence

behaviors and 3 additional self-care behaviors) that were hypothesized to apply to a majority

of individuals living with vasculitis: 1) adherence to self-administered medication, 2)

adherence to medication administered by a health professional, 3) adherence to

recommended appointments with health professionals, 4) adherence to recommended

medical tests and immunizations, 5) adherence to recommended infection avoidance

behaviors, 6) dietary adherence, 7) exercise adherence, 8) adherence to symptom monitoring

recommendations, 9) adjusting one's activity level based on current symptoms and fatigue,

10) prompt reporting of new or increased illness symptoms to a health professional, and 11)

prompt reporting of new or increased treatment side effects to a health professional. Within

each of the eight adherence sections, patients were first asked if a health professional had

recommended that they perform the particular type of behavior (e.g., taking medication), and

then to provide more details about the specific behavioral recommendations (e.g., how many,

when, and how medications are supposed to be taken). If patients had not been asked to

perform the type of behavior, they were instructed to skip to the next section of the

questionnaire. Patients who had been asked to perform the behavior were then asked follow-

up questions about how frequently they followed the recommendation(s) in recent weeks

and/or months. Two different recall periods were used, depending on the frequency with

which patients are typically required to perform the behavior. For behaviors that are typically performed on a daily basis (e.g., medication-taking, following dietary guidelines), patients were asked to report the frequency with which they performed the behavior according to their physicians' recommendations in the past four weeks. This recall period was thought to be long enough to produce sufficient opportunity for non-adherence and in turn, variation in responses among respondents, but short enough to produce reliable and accurate self-reports of behavior. For behaviors that are performed with varying frequency and/or much less often than daily (e.g., attending appointments) patients were asked to report their behavior during the past year. The response format varies across items in the VSMS, but consistently uses five-point Likert scales (e.g., *none of the time* to *all of the time*). Several items pertaining to each adherence behavior are negatively worded and reverse-scored, so that higher scores on each item correspond to higher levels of adherence. The format of the sections that assess the remaining three self-care behaviors (adjusting activity level in response to fatigue or symptoms, reporting illness symptoms, and reporting side effects) is very similar to the format of the adherence section. The only exception is that patients only skipped these sections if they had not experienced any fatigue or symptoms during the recall period. That is, the skip pattern is not based on their physicians' recommendations, because our conversations with ANCA-SVV health professionals and patients revealed that these self-care behaviors are appropriate for all ANCA-SVV patients when they experience fatigue or illness symptoms.

   The initial testing of the measure resulted in the deletion of the section about medication administered by a health professional, because approximately 75% of respondents did not report being prescribed any such medication in the past year and as a result, there were not

enough complete data in this section for analysis. In addition, factor analysis revealed that items pertaining to prompt reporting of illness symptoms and prompt reporting of side effects represented one common factor, and that items pertaining to adherence to recommended appointments with health professionals and adherence to recommended medical tests and immunizations also represented one common factor. Thus, the scoring of the final VSMS results in a total of eight subscale scores representing individuals' performance of eight self-management behaviors: 1) self-administered medication adherence, 2) adherence to recommended health services, 3) adherence to infection avoidance recommendations, 4) dietary adherence, 5) exercise adherence, 6) adherence to symptom monitoring recommendations, 7) adjusting activity level in response to fatigue or symptoms, and 8) prompt reporting of new or increased illness symptoms or side effects to a health professional. These eight self-management subscale scores were used as dependent variables in the current study.

Perceived Barriers to Performing Specific Self-management Behaviors

Within each of the original 11 VSMS sections that assessed performance of ANCA-SVV self-management behaviors, an additional item assessing patients' perceived barriers to performing the behavior in question was included, immediately following the items assessing level of adherence or self-care for that behavior. This item consisted of a checklist of factors and situations that could potentially prevent one from carrying out the self-management behavior. Patients were first assured in the instructions for this item that *"there are many factors that can get in the way of"* performing the self-management behavior, and then were asked to *"please check all of the factors that most frequently or consistently got in the way of"* performing the behavior during the recall period. The content of each checklist was

developed using data from semi-structured pilot interviews conducted with 18 adults living with ANCA-SVV as part of the development of the VSMS (see Thorpe *et al.*, in preparation for more details), and, thus, the number and nature of potential barriers included in each checklist varied for each self-management behavior (range=8-20 barriers per checklist). Respondents were also permitted to check "other" and write in additional barriers not included in the checklist. If respondents did not perceive any barriers to performing the self-management behavior, they could check *"not applicable – it was always easy for me to (perform the behavior)."* Dichotomous variables were then created for each barrier included in the checklist, as well as for additional barriers listed under "other" barriers. Each of these dichotomous variables was scored 1 if the barrier was checked or listed under other barriers, and 0 if not. In addition, a summary variable was created to reflect the total number of barriers endorsed by each respondent for each behavior.

Perceived Facilitators to Performing Specific Self-management Behaviors

Within each of the 11 VSMS sections and immediately following the barriers checklist, respondents were asked, using an open-ended question, to report perceived facilitators to performing the self-management behavior in question. The instructions for this item asked respondents, *"In the space below, please list the factors that you feel played the most important role in helping you to"* perform the specific self-management behavior during the recall period. Participants' responses to these open-ended items were then classified into categories of facilitators (e.g., utilization of organization tools such as calendars, logs, or pillboxes to help with medication-taking), and dichotomous variables were then created for each of these facilitators listed. As with the barriers items, each of these dichotomous variables was scored 1 if the facilitator was listed by the respondent and 0 if not. In addition,

a summary variable was created to reflect the total number of facilitators listed by each respondent for each behavior.

Perceived Difficulty of Specific Self-management Behaviors

Also included in each of the 11 self-management sections of the original VSMS was an item assessing respondents' perceived difficulty in performing the specific self-management behavior in question. Respondents were asked to rate on a 6-point Likert scale how difficult it is for them to perform the behavior exactly as recommended (*1=not at all difficult* to *6=extremely difficult*). These items were pilot-tested along with the rest of the VSMS, and demonstrated comprehensibility and variation in responses among respondents.

Perceived Importance of Specific Self-management Behaviors

A similar item was included in each of the 11 self-management sections of the original VSMS to assess respondents' perceptions of the importance of carrying out the specific self-management behavior. Respondents were asked to rate on a 6-point Likert scale in general, how important it is for them to perform the behavior exactly as recommended (*1=not at all important* to *6=extremely important*). These items were also pilot-tested along with the rest of the VSMS, and demonstrated comprehensibility and variation in responses.

Socio-demographics

In addition to completing the VSMS, patients reported their age (in years), gender, race/ethnicity, education (in years), and marital status.

Clinical and Health Variables

A number of clinical and health variables were also assessed to further characterize the patient sample. Patients were asked to report any other major medical diagnoses and their history, if any, of dialysis and kidney transplant. Illness duration, in months, was assessed by asking participants to report the month and year of diagnosis with ANCA-SVV, and then subtracting these values from the month and year of questionnaire completion. Current disease activity was assessed by both physician and patient report. Patients were asked to rate on a scale from 1 (not active at all/remission) to 10 (extremely active) how active their vasculitis currently was. Physicians were also asked to use the same rating scale to assess the patient's disease activity. Morbidity and damage from vasculitis were assessed via physician report using a similar 10-point scale (1=no damage to 10=extreme damage). Current health status was assessed using the General Health Scale of the SF-36, Version 2.0, scored using the RAND method (J. E. Ware, Jr. & Sherbourne, 1992).

Social Desirability Bias

Social desirability, or the desire of respondents to appear in ways that society regards as positive, was assessed in the study questionnaire using the short form of the Marlowe-Crowne Social Desirability Scale (Strahan & Gerbasi, 1972). This scale is widely used in psychometric studies to determine whether answers to self-reported questionnaires are influenced by respondents' desires to appear in socially desired ways. Cronbach's $\acute{a}$ in this study was .77.

<center>*Analysis*</center>

All analyses were conducted using SAS Version 9.13 (SAS Institute, Inc., Cary, NC).

Descriptive Analyses

To characterize the patient sample, descriptive statistics for all socio-demographics, clinical, and health variables described above were first examined. To further characterize the behavioral regimens recommended to respondents by health care providers, descriptive statistics were generated for the items assessing specific details of the behavioral recommendations that respondents reported having received from their health care providers (e.g., which specific types of medical tests and immunizations were recommended, exact nature of exercise recommendations). Next, for each of the 11 self-management behaviors assessed by the original VSMS, summary statistics were generated for the perceived difficulty ratings, the total number of barriers endorsed, perceived importance ratings, and the total number of facilitators listed. Finally, for each of the 11 behaviors, the frequencies of specific barriers and facilitators endorsed or listed were generated to identify those listed by at least 10% of the study sample.

Regression Models Predicting Self-management Behavior

To assess whether overall attitudes and beliefs about self-management behaviors predicted patients' report of their performance of these behaviors, a series of regression analyses were conducted. Separate regression models were evaluated for each of the self-management behaviors. Each of these models included the four primary behavior-specific independent variables (perceived difficulty of the behavior, the total number of barriers endorsed for the behavior, perceived importance of the behavior, and the total number of facilitators listed for the behavior), along with eight additional variables as controls (gender, race/ethnicity, age, years of education, marital status, duration of ANCA-SVV diagnosis, patient ratings of disease activity, and social desirability bias). It should be noted that for the VSMS subscale

of adherence to recommended health services, overall attitudes and beliefs about attending

recommended appointments with health professionals as well as attitudes and beliefs about

obtaining medical tests and immunizations (separately assessed in the original VSMS) were

analyzed as independent variables in separate regression models. This was deemed

appropriate given that the VSMS factor of adherence to recommended health services was

found to capture both appointments with health professionals and obtaining tests and

immunizations. Likewise, for the VSMS subscale of prompt reporting of illness symptoms

and side effects, overall attitudes and beliefs about reporting illness symptoms as well as

attitudes and beliefs about reporting treatment side effects (again, separately assessed in the

original VSMS) were analyzed as independent variables in separate regression models.

Again, this was considered appropriate given that reporting of illness symptoms and side

effects represent one self-management factor. Thus, a total of ten regression models were

examined in this set of analyses.

Because this set of analyses involved the evaluation of 40 regression coefficients (i.e., 10

models with 4 attitude/belief variables per model=40 regression coefficients of interest), we

used the Benjamini-Hochberg procedure for controlling the false discovery rate when

conducting multiple comparisons (Benjamini & Hochberg, 1995; Thissen, Steinberg, &

Kuang, 2002). The false discovery rate (FDR) is defined as the expected proportion of false

rejections of the null hypotheses for a set of planned comparisons, and differs from the

family-wise Type I error (FWE), which is the chance of *any* false rejections for the set of

comparisons (Keselman, Cribbie, & Holland, 2002). Controlling the FDR using the

Benjamini-Hochberg procedure has substantially more statistical power than the very

conservative, widely used methods of controlling the FWE using procedures such as the

Bonferroni correction, but still controls the expected proportion of false rejections for the set of comparisons to less than ά/2 (Keselman, *et al*., 2002; Thissen, *et al*., 2002). The Benjamini-Hochberg procedure is particularly recommended for exploratory research such as the current study, when a few Type I errors may be acceptable given the lack of prior research on the topic (Keselman, *et al*., 2002). We followed instructions for implementing the Benjamini-Hochberg procedure as outlined by Thissen and colleagues (2002). In brief, the *p*-values associated with the 40 beta coefficients were listed in ascending order and compared to linearly interpolated critical values ranging from ά/2 to ά/2/*m* (where *m*=the total number of comparisons, equal to 40 in this case). Observed *p*-values that were smaller than these corresponding critical values were deemed statistically significant.

We then conducted another series of regression analyses to assess whether perceptions of specific barriers and facilitators frequently reported by patients were associated with self-management behavior. For these analyses, we only evaluated effects of barriers and facilitators that were endorsed by at least 10% of the sample for which the behavior had been recommended. Each model consisted of one of the eight self-management scores as the dependent variable, a specific barrier or facilitator as the main independent variable, and the eight control variables listed above. We used the Benjamini-Hochberg procedure to evaluate the statistical significance of each test with the set of regressions examining specific barriers as predictors of self-management behavior (*m*=55), and again in the set of regressions examining specific facilitators (*m*=25).

For all regression analyses, we explored the effect of outliers on the results. We calculated Cook's *D* for each regression equation, to identify the individual observations which had a large influence on the regression coefficients produced by the model simultaneously (Kohler

& Kreuter, 2005). We then re-ran each regression equation, deleting those observations with a Cook's *D* greater than 4/*n* (Kohler & Kreuter, 2005) and re-evaluated the p-values within the set of regression equations according to the Benjamini-Hochberg procedure. Results that changed substantively after deleting outliers according to this procedure are presented along with the results of the regression equations before deleting outliers.

<div align="center">Results</div>

<div align="center">*Descriptive Results*</div>

Sample Characteristics

Descriptive statistics for the study sample, including socio-demographics and clinical variables, are shown in Table 5.1. Just over half of the sample was female, and almost all respondents reported being White/Caucasian. The mean age of the sample was 54.7 years, and the mean level of education was 14.6 years, or some college. The majority of the sample reported a diagnosis of Wegener's granulomatosis, and the mean duration of living with ANCA-SVV was 75.6 months, or just over six years (median=52.0 months, or 4.3 years). The average score on the General Health scale of the SF-36 was 45.0, which is substantially lower than the mean of 71.9 reported for the general U.S. population (J.E. Ware, Jr., 2000), but similar to that reported for patients with rheumatoid arthritis (*M*=44, *SD*=23; Ruta, *et al*., 1998).   The socio-demographic profile of this sample is similar to those previously reported for other United States samples of adults with systemic vasculitis (Carruthers, *et al.,* 1996; Hoffman, *et al.,* 1998).

Characteristics of Behavioral Recommendations

Patients had been recommended, on average, six of the eight possible adherence behaviors assessed by the VSMS. A summary of the specific behavioral recommendations received by the patient sample from health care providers is shown in Table 5.2. Almost all patients had been instructed to take self-administered medication during the past four weeks, to attend appointments with health professionals in the past year, and to obtain immunizations or medical tests in the past year. Most respondents reported being prescribed medication to put or keep their vasculitis in remission as well as medication to control complications of vasculitis or its treatment. Patients were taking an average of 6.8 (SD=4.1) prescribed medications for reasons related to ANCA-SVV (not shown in table). Nephrologists, primary care physicians, opthamologists, and rheumatologists were the most common types of health professionals that patients were instructed to see. The mean number of health professionals that patients had been recommended to see in the past year was 4.4 (SD=2.0, range 0-11, not shown in table). While several types of medical tests and immunizations were relatively common, the flu shot was recommended to most patients.

In addition, a majority of patients had been instructed to take steps to avoid getting an infection, follow dietary recommendations, follow exercise recommendations, and monitor illness symptoms in the past four weeks. The most common infection avoidance behaviors recommended to patients included staying away from people who are ill, washing hands frequently, and avoiding crowds. While dietary recommendations represented a wide range of specific behaviors, the most common recommendations included following a low-salt diet, limiting alcohol intake, trying to lose weight and following a low-cholesterol diet. Among exercise recommendations, cardiovascular exercise was most commonly recommended,

although these recommendations were often general in nature (e.g., get some exercise, walk most days of the week). Among those that reported having received a specific exercise recommendation, instructions were quite varied with regard to time spent in exercise and number of exercise sessions per week (e.g., twice a week for 20 minutes a session to 7 days a week for 60 minutes a session). Among symptom monitoring tasks, at-home weight and blood pressure monitoring were most common. The only behavior assessed by the original VSMS that was not frequently recommended to patients in this sample was taking medication administered in a doctor's office or clinic.

The sections of the VSMS assessing additional self-care behaviors not necessarily recommended by health care providers but generally recommended for all ANCA-SVV patients were applicable for and completed by the vast majority of respondents. Over 90% of patients reported feeling tired, ill, or run-down during the past four weeks at least "*a little of the time*" and, thus, completed the items on adjusting activity level in response to fatigue or symptoms (not shown in table). Approximately 94% and 86.8% of respondents completed the items on reporting illness symptoms and side effects, respectively (not shown in table).

Level of Performance of Self-management Behaviors

Descriptive statistics (*n, M, SD,* and Cronbach's *ά*) for the eight VSMS subscales are shown in Table 5.3. The highest levels of self-management were found for adherence to recommended health services and medication adherence, while the lowest levels were found for exercise adherence, prompt reporting of symptoms and side effects, and adjusting activity level in response to fatigue and symptoms.

Perceptions of Barriers and Facilitators

Summary statistics describing perceived difficulty ratings and the mean number of barriers endorsed by patients for each of the 11 self-management behaviors are shown in the second and third columns of Table 5.4. Patients rated medication-taking (assessed globally for all medications regardless of whether they were self-administered or health professional-administered) lowest with regard to perceived difficulty. In addition, patients, on average, endorsed the fewest number of barriers for health-professional-administered medication, obtaining tests and immunizations, attending appointments with health professionals, and symptom monitoring adherence. Exercise, diet, and adjusting activity level in response to fatigue or symptoms received the highest perceived difficulty ratings. On average, patients also endorsed the greatest number of barriers for exercise. In addition, patients reported a mean of more than two barriers for reporting symptoms, reporting side effects, diet, and adjusting activity level in response to fatigue or symptoms. Within each self-management behavior, perceived difficulty and the total number of barriers tended to be moderately to strongly and positively correlated with one another (r's ranging from 0.38 to 0.60). The remaining bivariable correlations among the four attitude and belief variables assessed for each self-management behavior tended to be relatively small (r's <.30).

Perceived importance ratings and mean number of facilitators listed by patients for each behavior are shown in the fourth and fifth columns of Table 5.4. Patients rated medication-taking, attending appointments, and obtaining tests and immunizations highest with regard to perceived importance. Patients, on average, listed the greatest number of facilitators for self-administered medication, attending appointments, and symptom monitoring adherence. Exercise received the lowest perceived importance ratings, followed closely by diet,

reporting symptoms, reporting side effects, and adjusting activity level in response to fatigue or symptoms. Patients reported the fewest number of facilitators for diet, reporting side effects, reporting symptoms, and infection avoidance.

Specific barriers and facilitators to performing each of the eight adherence behaviors and three self-care behaviors that were reported by at least 10% of patients are shown in the first and second columns of Table 5.5 and Table 5.6, respectively. These barriers and facilitators are discussed in more detail below with the results of the regression models.

Regression Models Predicting Self-management Behavior

Relationship of Overall Attitudes and Beliefs to Self-management

The results of the ten regression models examining the relationship of perceived difficulty ratings, perceived importance ratings, total number of perceived barriers, and total number of perceived facilitators to each of the eight self-management behaviors are shown in Table 5.7. Perceived difficulty ratings of specific behaviors were independently and negatively associated with patients' reports of their performance of that behavior, across all eight self-management behaviors assessed by the VSMS, with one exception: perceived difficulty of attending appointments with health professionals was not associated with adherence to recommended health services, after all other variables were controlled. In addition, the total number of barriers endorsed was significantly and negatively associated with performance of each behavior, with the exception of symptom monitoring adherence. Also as expected, perceived importance of each self-management behavior positively predicted performance of the behavior, with one exception: perceived importance of attending appointments with health professionals was not associated with adherence to recommended health services, after all other variables were controlled. Contrary to expectations, the relationship between the

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total number of facilitators listed by patients and performance of self-management behaviors was not statistically significant, with one exception. Patients who reported a greater number of perceived facilitators to exercise adherence tended to have higher levels of exercise adherence. None of the results shown in Table 5.7 changed substantively after deleting observations with Cook's D greater than $4/n$.

Relationship of Specific Barriers to Self-management

The results of the regression equations examining specific perceived barriers to performance of the eight specific behaviors as predictors of actual self-management behavior are shown in Table 5.5.

Endorsement of all five perceived barriers frequently mentioned by patients with regard to self-administered medication adherence was significantly and negatively associated with adherence. The first two of these barriers, mentioned by over 30% of patients who had been prescribed self-administered medication, represented disruptions to patients' daily routines and simply forgetting to take the medication. The remaining three factors represented factors pertaining to the complexity of the medication regimen.

Only one barrier to taking medication administered by a health professional was mentioned by at least 10% of individuals who had been prescribed such medication: having to travel a long way to get the medication. As explained above, we were not able to look at the association between endorsement of this barrier and adherence, because the VSMS does not measure adherence to health professional-administered medication.

Of the frequently reported barriers to attending recommended appointments with health professionals, none were significantly associated with adherence to recommended health services. Likewise, the only perceived barrier to obtaining tests and immunizations that was

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frequently mentioned by patients, trouble scheduling the appointment at a time that was convenient, was not significantly associated with adherence to recommended health services.

For adherence to infection avoidance recommendations, one of the five perceived barriers that were frequently mentioned by patients was significantly and negatively associated with adherence: lower levels of infection avoidance behavior were reported by those patients endorsing the barrier of work responsibilities getting in the way (b=-.69, se=.15, p<.0001).

Four of the six frequently mentioned perceived barriers to dietary adherence were significantly and negatively associated with dietary adherence. That is, lower levels of dietary adherence were reported by patients endorsing the following barriers: often being around foods they are not supposed to eat (b=-.42, se=.13, p=.0012), having dietary recommendations that required them to eat differently than they prefer to eat (b=-.58, se=.13, p<.0001), a belief that emotions caused them to seek comfort in foods they are supposed to avoid (b=-.65, se=.15, p<.0001), and low motivation to follow the recommendations (b=-.56, se=.16, p=.0006).

For exercise adherence, endorsement of five of the 11 perceived barriers reported by at least 10% of patients receiving exercise recommendations were significantly associated with worse adherence. These barriers included not having enough energy to exercise (b=-.49, se=.20, p=.0142), not feeling motivated to exercise (b=-1.00, se=.17, p<.0001), a belief that work responsibilities got in the way (b=-.78, se=.23, p=.0011), a belief that family responsibilities got in the way (b=-.69, se=.24, p=.0044), disliking exercise (b=-1.18, se=.25, p<.0001), and not having anyone with whom to exercise (b=-.78, se=.32, p=.0208).

For adherence to symptom monitoring recommendations, endorsement of two of the four frequently perceived barriers was significantly and negatively associated with adherence.

These barriers included simply forgetting (b=-.99, se=.14, p<.0001) and a belief that the behavior was not necessary for controlling their symptoms (b=-1.13, se=.25, p<.0001).

Reporting of four of seven frequently endorsed perceived barriers was significantly and negatively associated with patients' appropriate adjusting of their activities in response to fatigue or illness symptoms. That is, patients who less frequently cut back on their activities when they felt tired, ill, or run-down were more likely to endorse the following barriers: family responsibilities got in the way (b=-.26, se=.10, p=.0089), work responsibilities got in the way (b=-.35, se=.11, p=.0013), not feeling comfortable with asking others to help with responsibilities (b=-.42, se=.11, p=.0001), and feeling that others would not understand limiting their activities (b=-.41, se=.11, p=.0004).

For prompt reporting of illness symptoms and side effects, endorsement of all frequently reported barriers predicted lower levels of symptom and side effect reporting within one day, with one exception (see Table 5.5). This held true for perceived barriers to reporting illness symptoms as well as perceived barriers to reporting side effects, many of which overlapped.

In this set of analyses, six additional beta coefficients became statistically significant after deleting observations with a Cook's D greater than 4/n from each analysis. These beta coefficients, all of which fell just short of statistical significance in the original models, are shown in Table 5.8.

Relationship of Specific Facilitators to Self-management

The results of the regression equations exploring the relationship of specific facilitators reported by patients to their self-reported performance of the corresponding self-management behaviors are shown in Table 5.6. Across all self-management behaviors, reporting of specific perceived facilitators was not significantly associated with performance of the

behaviors, although many of the p-values fell just short of statistical significance. These results did not change substantively after deleting observations with a Cook's *D* greater than 4/*n* from each analysis.

Discussion

To our knowledge, this is the first study to examine patients' attitudes and beliefs about their illness and its treatment to self-management behavior among adults living with ANCA-SVV. In addition to characterizing patients' overall perceptions of the difficulty and importance of different self-management behaviors, we identified specific perceived barriers and facilitators that patients frequently experience with regard to each behavior. Furthermore, we found that patients' perceptions regarding the difficulty and importance of various self-management behaviors, and the reporting of several specific perceived barriers, were significantly associated with patients' self-management behavior.

Our results suggest that patients believe that among the self-management challenges they face, they experience the least difficulty in using recommended health services and taking medication. Patients reported the highest levels of adherence, the highest levels of perceived importance, and greatest number of perceived facilitators for these behaviors. They also rated these behaviors lowest with regard to perceived difficulty. In contrast, analysis of these same indicators revealed that patients appeared to have the most difficulty with exercise adherence, adjusting activity level in response to fatigue or symptoms, and prompt reporting of symptoms and side effects. Patients also perceived dietary recommendations as difficult to follow, as demonstrated by high perceived difficulty ratings, a high average total number of reported barriers, and a low average total number of reported facilitators, and less important to follow, although mean levels of actual dietary adherence were more moderate.

These findings are not surprising, given that prior research with other illness populations has shown that patients are typically less adherent to self-management behaviors involving lifestyle modifications, such as diet and exercise (Bosworth et al., 2006; Johnson, 1992; Pham *et al.*, 1996)

   As expected, patients' overall perceptions of barriers, as indicated by perceived difficulty ratings and the average number of specific barriers endorsed, were significantly associated with self-management behavior, with just a few exceptions. That is, patients who perceived behaviors as more difficult to perform and who reported a greater number of specific barriers to performing the behavior also tended to report lower levels of adherence and self-care, independent of perceived importance and the total number of perceived facilitators. Furthermore, with one exception, the more important patients perceived a specific self-management behavior to be, the more frequently they reported performing it as recommended. While the total number of reported facilitators to carrying out exercise recommendations was associated with exercise adherence, the total number of reported behavior-specific facilitators did not predict performance of the behavior after controlling for other important variables. These results, taken with prior findings that perceived barriers most strongly predict health behavior compared to other perceptions, such as perceived benefits (Janz & Becker, 1984), suggest that patients' perceptions of barriers may play a more important role than perceived facilitators in determining self-management behavior in this population. However, we cannot rule out that differences in the measurement of perceived barriers and perceived facilitators are responsible for the differential observed relationships; i.e., the greater response burden required by patients in generating and listing facilitators compared to selecting from a list of barriers may have introduced more

measurement error for perceived facilitators, in turn attenuating its relationship with self-reported behavior.

This research also identified some of the specific perceived barriers and facilitators that are most salient to the patients studied and that may also affect actual self-management behavior. Across behaviors, perceived benefits (e.g., a belief that performing the behavior is very important for maintaining good health) were frequently mentioned as facilitators for many of the self-management behaviors assessed in this study. It is interesting to note that perceiving benefits of the behavior as a facilitator to carrying out the behavior was not associated with higher levels of self-management. As suggested by a vast body of previous health behavior theory and research, as well as research on the health belief model specifically (Janz & Becker, 1984), perceiving benefits of the behavior may be helpful, but not sufficient, for performing the behavior as recommended.

With the exception of perceived benefits, the barriers and facilitators most frequently mentioned by patients varied substantially across behaviors, as did significant relationships between reporting of these barriers and self-management behavior. This provides further support for the argument that each self-management behavior is distinct from one another, and efforts to improve patients' level of self-management should be behavior-specific.

Within each self-management behavior assessed systematically by the VSMS in this study, patients' reporting of a number of specific barriers was found to significantly predict their level of performance of the behavior. These perceived barriers may be especially fruitful avenues to explore in future research and intervention with ANCA-SVV patients. While space limitations prohibit an exhaustive discussion of each of these factors, a few findings warrant highlighting here.

Among the self-management behaviors that have been extensively studied with other patient populations, including medication adherence, adherence to recommended health services, dietary adherence, and exercise adherence, many of the most frequently reported barriers and facilitators in this study have been previously documented as influences on adherence. For example, routinization of one's daily activities, and incorporating medication-taking into this routine, have been previously found to facilitate medication adherence among individuals living with HIV (Ryan & Wagner, 2003; Wagner & Ryan, 2004), a disease in which medication regimens can also be complex. Similarly, the use of pillboxes for organizing medication regimens has been associated with improved medication adherence among individuals living with HIV (Kalichman *et al.*, 2005). As with prior research in other rheumatic disease populations (Garcia Popa-Lisseanu *et al.*, 2005), a high proportion of patients in this study also indicated that the difficulty or ease with which they could schedule appointments at preferred times played a key role in their ability to adhere to recommendations for using health services, although reporting of this as a barrier or facilitator was not related to self-reported adherence. Although not significantly related to adherence to recommended health services in this study, geographic distance from health care providers was also frequently mentioned as a barrier to attending appointments with health professionals, just as greater geographic distance from health care facilities has been documented as a negative influence on utilization of outpatient care in other patient populations (McCarthy & Blow, 2004). Likewise, the specific barriers mentioned by patients and associated with worse dietary adherence, namely frequent exposure to prohibited foods, food preferences, emotional eating, and low motivation to adhere, have been found to be powerful predictors of dietary behavior in other patient populations (Schultz *et al.*, 2001;

Yancy & Boan, 2006). Finally, lack of motivation, time, facilities, and someone with whom

to exercise have all been associated with reduced physical activity levels in prior research

(Dominick & Morey, 2006) and are reflected in this study as well. The similarities of the

perceived barriers to these specific self-management behaviors in ANCA-SVV patients

compared to other chronically ill individuals suggests that researchers and clinicians may

want to look to interventions that have been used successfully with other patients for

guidance in developing intervention strategies for ANCA-SVV patients for improving these

self-management behaviors.

Much less research has been conducted on the remaining four self-management behaviors

assessed in this study: adherence to recommended infection avoidance behaviors, symptom

monitoring adherence, adjusting activity level in response to fatigue or symptoms, and

prompt reporting of illness symptoms and side effects. In this study, the barriers reported for

adhering to recommended infection avoidance behaviors revealed that patients' desire to

carry on with their normal activities, whether they are work-related, family-related, or

social/leisure-related, were perceived to conflict with their adherence, although only the

belief that work responsibilities got in the way was significantly related to adherence. With

regard to adjusting activity level in response to fatigue or symptoms, respondents once again

commonly perceived that their responsibilities, both work and family related, made it

difficult to obtain the rest their bodies needed; on the flip side, others perceived reduced or

flexible work schedules and retirement as a key facilitator. Many respondents also reported

reluctance in reducing their activities or asking for help because of worries about others'

reactions to them doing so. Given that previous research has found that a substantial

proportion of individuals with ANCA-SVV experience disruptions at work, reductions in

household income, and negative influences on interpersonal relationships due to their illness (Hoffman *et al.*, 1998), it is not surprising that some patients are hesitant to reduce their activity levels or delegate their responsibilities to others despite signals from their bodies that they should, or alter their work, family, or leisure activities to attempt to avoid getting an infection. These results suggest that with regard to these behaviors, some patients appear to view the costs of strictly adhering to self-care guidelines as outweighing the benefits.

With regard to symptom monitoring adherence, simple forgetting and low perceived benefits (i.e., a belief that symptom monitoring was not necessary for controlling one's condition) were commonly reported, and reporting of these perceptions predicted worse adherence. Finally, prompt reporting of new or increased illness symptoms and side effects appears to be a particularly difficult issue for many patients, as demonstrated by the frequency with which a number of barriers to this action were endorsed by patients. Most notably, patients reported that their uncertainty as to whether symptoms are related to vasculitis or its treatment or are meaningful enough to report often served as barriers to prompt reporting. Many patients were wary of going through the trouble of trying to speak with a health professional or "bothering" the health professional about something that might not be important, and were inclined to wait and see if the symptom would resolve on its own. Endorsement of these barriers was in turn related to greater delay in reporting of symptoms and side effects. Future studies should further explore the role of the patient-physician relationship, and specific provider behaviors, as barriers or facilitators to prompt reporting of symptoms and side effects by ANCA-SVV patients.

Several limitations of this study should be noted. First, this study was cross-sectional and thus limits our ability to draw causal inferences about the significant relationships observed

in this study. Second, we used a convenience sample of ANCA-SVV patients that may not represent the larger population of ANCA-SVV patients. In particular, because we used a registry of patients with autoimmune kidney-related diseases as a primary recruitment source, our sample may have over-represented patients with kidney involvement. Furthermore, the average disease duration reported by this sample was over six years, and we may have under-represented recently diagnosed individuals. Third, our measurement of perceived barriers and in particular, perceived facilitators, was less than ideal in that neither used established, validated measures and all barriers and facilitators were assessed using single items. Furthermore, as mentioned above, the open-ended items assessing perceived facilitators placed a high burden on respondents. As a result, respondents may not have listed all the relevant facilitators they experience with regard to illness self-management behaviors. Future research should examine the perceived barriers and facilitators identified in this research to be important to self-management using more established measures. Fourth, this study was limited to assessing patients' *perceptions* of barriers and facilitators, and should by no means be considered an exhaustive study of factors that impede or support ANCA-SVV self-management. Respondents may not explicitly recognize the factors that influence their behavior. Fifth, we also relied on patient recall for assessing which self-care behaviors had been explicitly recommended to them by a health professional; thus, the six adherence subscales are actually measuring *perceived* adherence. As a result, the relationships between self-management behavior and patients' attitudes and beliefs may have been inflated due to the self-report measurement used to assess both types of variables. In addition, some patients may have failed to recall all of their physicians' recommendations and may have skipped items pertaining to these behaviors. This could have led to non-adherent individuals being

more likely to be excluded from each behavior-specific analyses, which would have in turn inhibited our ability to detect significant relationships between the key independent variables and self-management behavior. Finally, there was considerable heterogeneity with regard to the specific behavioral recommendations that patients reported receiving from health professionals within each behavioral domain (e.g., some patients' dietary recommendations consisted only of limiting alcohol intake, while others were asked to restrict intake of multiple nutrients), and we were not able to explore whether variation in behavioral recommendations was associated with variation in perceived barriers and facilitators and/or adherence.

Despite these limitations, this study provides some insight into ANCA-SVV patients' perceived barriers and facilitators to performing a number of self-management behaviors, as well as evidence that perceived barriers and facilitators may influence self-management in this patient population, just as in other patient populations. The findings in this study serve as an excellent starting point for further study into important influences on illness self-management in this illness context, including the development and testing of intervention strategies.

Table 5.1. Descriptive statistics for ANCA-SVV patients (n=205).

	n	*% or M (SD)*
Socio-demographics		
Gender		
Male	95	46.3%
Female	110	53.7%
Missing	0	0%
Race/ethnicity		
White	191	93.2%
American Indian/Alaskan Native	2	1.0%
Asian	5	2.4%
Black or African American	2	1.0%
Hispanic/Latino	4	2.0%
Missing	1	0.5%
Age, in years	205	54.7 (14.7)
Years of education	205	14.6 (2.4)
Marital status		
Married	152	74.2%
Non-married	53	25.9%
Missing	0	0%
Clinical characteristics		
Self-reported condition		
Wegener's granulomatosis	145	70.7%
Microscopic polyangiitis	16	7.8%

Churg Strauss syndrome	10	4.9%
ANCA-glomerulonephritis	31	15.1%
Good Pasture's disease	2	1.0%
Temporal arteritis	1	0.5%
Missing	0	0%
Time since diagnosis, in months	204	75.7 (70.7)
Time between symptom onset and diagnosis, in months	202	13.9 (34.0)
Patient-perceived disease activity, range 1-10	204	2.9 (2.1)
Physician-perceived disease activity, range 1-10	143	1.9 (1.3)
Physician-perceived damage, range 1-10	119	4.4 (2.6)
General physical health, range 0-100	205	45.0 (23.6)
Ever on dialysis		
Yes	32	15.8%
No	170	82.9%
Missing	3	1.5%
History of kidney transplant		
Yes	15	7.3%
No	186	90.7%
Missing	4	2.0%

Table 5.2. Summary of behavioral recommendations received by ANCA-SVV patients from health care providers.

Behavioral Recommendation	%	n
Take self-administered medication (past 4 weeks)		
Any medication for reasons related to vasculitis, *%, n*	95.1%	195
Medication to put or keep vasculitis in remission, *%, n*	83.4%	171
Medication to control complications due to vasculitis or its treatment, *%, n*	87.8%	180
Take medication administered in a doctor's office or clinic (past year)		
Any medication	25.4%	52
Attend appointments with health professionals (past year)		
Any health professional	99.0%	203
Rheumatologist	51.5%	104
Nephrologist	67.7%	138
Pulmonologist	32.5%	66
Ear, nose, and throat specialist (ENT)	39.7%	81
Opthamologist	53.4%	109
Urologist	27.7%	56
Oncologist	6.9%	14
Dermatologist	28.4%	58
Primary care physician	67.2%	137
Obstetrician/gynecologist	25.1%	51
Physical therapist	9.4%	19

Other (cardiologist, endocrinologist, GI, neurologist, dentist, hematologist, immunologist, infectious disease, periodontist, podiatrist, psychiatrist, radiologist, transplant surgeon, vasculitis clinic, counselor, surgeon; all n<8)	24.1%	49

Obtain immunizations and medical tests (past year)

Any immunization or test	99.0%	203
Flu shot	87.3%	179
Pneumonia shot	40.0%	78
Blood draws outside of regularly scheduled appointments	69.3%	142
Bone density test	51.5%	105
X-rays, CAT scans, MRIs, or other types of scans	69.1%	141
Other test (represented a wide variety of tests)	42.6%	87

Take steps to avoid getting an infection (past 4 weeks)

Any steps	70.1%	143
Avoid crowds	40.5%	83
Avoid shaking hands with people	26.3%	54
Wear a mask when out in public	7.8%	16
Carry and use hand sanitizer	29.3%	60
Wash hands frequently	61.5%	126
Stay away from people who are ill	64.4%	132
Avoid eating certain foods (e.g., restaurant-prepared foods, buffets, fresh vegetables)	8.8%	18
Other steps (represented a wide variety of steps, often more specific instructions to the above)	20.0%	41

Follow dietary recommendations (past 4 weeks)

Any dietary recommendations	78.1%	160
Try to gain weight	4.4%	9
Try to lose weight	36.6%	75
Follow a high-protein diet	8.3%	17
Follow a low-protein diet	8.8%	18
Follow a low-fat diet	24.4%	50
Follow a low-carbohydrate diet	10.7%	22
Follow a low-cholesterol diet	31.7%	65
Follow a low-salt diet	41.0%	84
Follow a low-potassium diet	14.6%	30
Follow a low-phosphorus diet	6.3%	13
Follow a low-magnesium diet	2.9%	6
Follow a diabetic diet	11.7%	24
Limit your alcohol intake	37.0%	76
Other dietary recommendation (represented a variety of recommendations)	9.3%	19

Follow exercise recommendations (past 4 weeks)

Any exercise recommendation	61.5%	126
Do planned cardiovascular activity	58.0%	119
Specific recommendation	36.2%	42
General recommendation	63.8%	74
Do planned strength-building exercise	22.9%	47
Specific recommendation	31.8%	14

General recommendation	68.2%	30
Do stretching, yoga, or other flexibility exercises	15.1%	31
Specific recommendation	34.6%	9
General recommendation	65.4%	17

Monitor illness symptoms (past 4 weeks)

Any monitoring recommendation	64.9%	133
Monitor weight	38.5%	79
Monitor blood pressure at home	42.9%	88
Monitor blood sugar	17.6%	36
Other monitoring task (testing urine at home most common, n=11)	11.7%	24

Table 5.3. Descriptive statistics for VSMS subscales.

	n	*M (SD)*	Cronbach's $\acute{a}$
Medication adherence	191	4.5 (.54)	.77
Adherence to recommended health services	202	4.6 (.63)	.78
Infection avoidance adherence	142	3.7 (.79)	.85
Diet adherence	154	3.6 (.76)	.76
Exercise adherence	125	3.2 (1.02)	.94
Symptom monitoring adherence	133	3.8 (.97)	.91
Adjusting activity in response to symptoms	185	3.4 (.64)	.67
Reporting symptoms and side effects	177	3.2 (1.08)	.90

*Range of all eight VSMS subscales was 1-5.

Table 5.4. Summary statistics for overall perceptions of barriers and facilitators to performing ANCA-SVV self-management behaviors.

	Perceived Difficulty (range=1-6) (*M, SD*)	Number of Barriers Endorsed (*M, SD)*	Perceived Importance (range=1-6) (*M, SD*)	Number of Facilitators Listed (*M, SD*)
Medication (administered by self and/or health professional)*	1.5 (1.0)*	1.5 (1.9)	5.9 (0.4)*	1.5 (1.0)
Appointments with health professionals	2.0 (1.4)	1.1 (1.6)	5.7 (0.7)	1.3 (0.9)
Tests and Immunizations	1.8 (1.3)	0.9 (1.4)	5.7 (0.6)	1.0 (0.8)
Adjusting activity level	3.0 (1.6)	2.2 (1.7)	4.8 (1.3)	1.2 (0.9)
Reporting symptoms	2.4 (1.5)	2.9 (2.3)	4.8 (1.4)	0.9 (0.8)
Reporting side effects	2.1 (1.5)	2.4 (2.1)	4.8 (1.5)	0.8 (0.8)
Infection avoidance	2.7 (1.7)	1.8 (1.4)	5.4 (0.8)	0.9 (0.7)
Diet	3.0 (1.5)	2.2 (2.0)	4.8 (1.2)	0.8 (0.9)
Exercise	3.2 (1.5)	3.1 (2.1)	4.7 (1.2)	1.0 (1.1)
Symptom monitoring	2.0 (1.3)	1.2 (1.2)	5.1 (1.1)	1.3 (1.3)

* Perceived importance and perceived difficulty were assessed globally, for all medication, regardless of mode of administration.

Table 5.5. Relationship of specific barriers to levels of eight self-management behaviors.

	% Endorsed	b	se	p
Self-administered Medication Adherence (n=187)				
1. My daily routine was disrupted.	31.8%	-.40	.08	<.0001*
2. I simply forgot to take it.	30.2%	-.59	.07	<.0001*
3. I take a large number of medications.	13.0%	-.29	.10	.0069*
4. The specific instructions (e.g., taking with meals, etc.) for taking medications are difficult to follow.	12.0%	-.42	.11	.0003*
5. My dosing schedule is complicated.	10.4%	-.43	.12	.0003*
Health Professional-administered Medication Adherence				
1. I had to travel a long way to get the medication.	11.5%	-	-	-
Adherence to Recommended Health Services (n=192) (barriers for attending appointments)				
1. Work responsibilities got in the way.	14.6%	-.06	.13	.65
2. I had trouble scheduling the appointment at a time that was convenient for me.	12.6%	-.09	.14	.54
3. I had to travel a long way to get there.	11.1%	-.30	.14	.035
Adherence to Recommended Health Services (n=198) (barriers for obtaining tests and immunizations)				
1. I had trouble scheduling the appointment at a time that was convenient for me.	11.9%	-.22	.14	.12
Adherence to Infection Avoidance Recommendations (n=140)				
1. My desire to participate in social/leisure activities got in the way.	40.9%	-.30	.14	.0312

2. It was impossible for me to avoid people who were sick.	36.6%	-.30	.14	.0428
3. Work responsibilities got in the way.	23.9%	-.69	.15	<.0001*
4. I simply forgot.	21.8%	-.35	.16	.0311
5. Family responsibilities got in the way.	19.0%	-.36	.18	.0438
Dietary adherence (n=151)				
1. I was often around foods I am not supposed to eat.	33.3%	-.42	.13	.0012*
2. My dietary recommendations required me to eat differently than I preferred to eat.	29.6%	-.58	.13	<.0001*
3. My emotions caused me to seek comfort in foods I am supposed to avoid.	21.4%	-.65	.15	<.0001*
4. My daily routine was disrupted.	20.8%	-.05	.15	.73
5. When eating outside the home, I did not have access to recommended foods.	19.5%	.03	.16	.83
6. I did not have the motivation.	17.0%	-.56	.16	.0006*
Exercise adherence (n=124)				
1. I did not have enough energy to exercise.	51.2%	-.49	.20	.0142*
2. I did not feel motivated to exercise.	48.8%	-1.00	.17	<.0001*
3. Bad weather prevented me from exercising outdoors.	31.2%	-.16	.21	.45
4. My daily routine was disrupted.	28.8%	.40	.21	.06
5. I felt too ill to exercise.	27.2%	-.24	.23	.30
6. Work responsibilities got in the way.	23.2%	-.78	.23	.0011*
7. Family responsibilities got in the way.	22.4%	-.69	.24	.0044*
8. I felt that I got enough exercise in my daily activities.	18.4%	-.43	.27	.11
9. I dislike exercising.	16.8%	-1.18	.25	<.0001*

10. I did not have access to things I needed to exercise.	13.6%	-.09	.28	.76
11. I did not have anyone to exercise with.	10.4%	-.78	.32	.0208

Adherence to Symptom Monitoring Recommendations (n=132)

1. I simply forgot.	47.4%	-.99	.14	<.0001*
2. My daily routine was disrupted.	26.3%	-.34	.20	.08
3. I did not think it was necessary for controlling my condition.	11.3%	-1.13	.25	<.0001*
3. Work responsibilities got in the way.	11.3%	-.51	.27	.06

Adjusting activity level in Response to Fatigue or Symptoms (n=182)

1. Family responsibilities got in the way.	38.4%	-.26	.10	.0089*
2. My desire to participate in social or leisure activities got in the way.	36.8%	-.09	.10	.34
3. Work responsibilities got in the way.	32.4%	-.35	.11	.0013*
4. I did not feel comfortable asking others to help with my responsibilities.	26.0%	-.42	.11	.0001*
5. I felt like others would not understand if I limited my activities.	23.4%	-.41	.11	.0004*
6. Medication side effects prevented me from resting or sleeping.	17.8%	-.22	.12	.07
7. Illness symptoms prevented me from resting or sleeping.	16.8%	-.07	.13	.59

Prompt reporting of Illness Symptoms and Side Effects (n=172) (barriers to reporting symptoms)

1. I thought the symptom might go away on its own.	65.6%	-.91	.16	<.0001*
2. I was not sure if the symptom was related to my vasculitis.	60.9%	-1.04	.16	<.0001*

3. I thought it could wait until my next appointment.	38.5%	-1.17	.14	<.0001*
4. I did not want to bother the health professional unnecessarily.	36.5%	-.93	.17	<.0001*
5. I was afraid of looking like a hypochondriac.	20.3%	-.41	.21	.05
6. It was difficult to reach a health professional.	19.3%	-.73	.20	.0003*
7. I did not want to be prescribed more medication as a result of reporting the symptoms.	11.5%	-.78	.25	.0023*
8. My doctor(s) have not recommended that I contact them within a day of noticing new/increased symptoms.	10.5%	-.77	.27	.0044*
Prompt reporting of Illness Symptoms and Side Effects (n=173) (barriers to reporting side effects)				
1. I thought the side effect might go away on its own.	56.2%	-.88	.15	<.0001*
2. I was not sure if the side effect was related to my vasculitis treatment.	48.3%	-.67	.16	<.0001*
3. I did not want to bother the health professional unnecessarily.	31.5%	-.85	.17	<.0001*
4. I thought it could wait until my next appointment.	30.3%	-.89	.17	<.0001*
5. It was difficult to reach a health professional.	16.9%	-.77	.21	.0004*

*p-value is significant, according to Benjamini-Hochberg procedure.

Table 5.6. Relationship of specific facilitators to levels of eight self-management behaviors.

	% Listed	b	se	p
Self-administered Medication Adherence (n=187)				
1. Organization tools (e.g., pillboxes, calendars).	27.1%	.17	.08	.0414
2. Establishing a routine.	19.8%	-.0003	.09	.99
3. Perceived benefits.	16.7%	.03	.10	.74
4. Plan ahead and anticipate routine disruptions.	14.1%	.27	.10	.0080
5. Take medications with other routine tasks.	12.5%	.08	.11	.43
Health Professional-administered Medication Adherence				
1. Positive health beliefs.	19.2%	-	-	-
2. Family support.	13.5%	-	-	-
3. Medication is administered at another doctor's appointment.	11.5%	-	-	-
Adherence to Recommended Health Services (n=192) (facilitators for attending appointments)				
1. Perceived benefits.	16.6%	.26	.12	.0324
2. Ease/flexibility in scheduling appointments.	11.6%	-.04	.14	.77
3. Good relationship with health care providers.	10.6%	-.14	.15	.33
3. Made it a priority and did not let inconvenience be an excuse.	10.6%	.02	.15	.89
Adherence to Recommended Health Services (n=198) (facilitators for obtaining tests and immunizations)				
1. Perceived benefits.	13.9%	.23	.13	.08
2. Convenient location of office.	12.4%	-.23	13	.07
Adherence to Infection Avoidance Recommendations (n=140)				

1. The participant listed specific measures and tricks that are helpful.	29.9%	-.06	.15	.68
2. Perceived benefits.	20.4%	.48	.18	.0056
Dietary adherence (n=151)				
1. Perceived benefits.	9.4%	.21	.21	.31
Exercise adherence (n=124)				
1. Perceived benefits.	12.0%	.40	.29	.18
2. Easy access to good workout facilities.	10.4%	.77	.30	.0122
2. Have a workout buddy (e.g., another person, dog).	10.4%	-.23	.32	.48
Adherence to Symptom Monitoring Recommendations (n=132)				
1. Having equipment or supplies on hand and easily accessible.	13.5%	-.22	.26	.39
2. Having an established routine (e.g., do it the same time each day).	12.0%	.63	.26	.0175
Adjusting activity level in Response to Fatigue or Symptoms (n=182)				
1. Support/influence from family.	27.0%	-.03	.11	.75
2. Being retired or reduced work schedule.	13.0%	.35	.14	.0126
Prompt reporting of Illness Symptoms and Side Effects (n=172) (facilitators for reporting symptoms)				
1. Having a doctor that is receptive.	17.2%	.37	.22	.09
2. Being able to reach doctor easily without a hassle.	16.7%	.06	.22	.77
Prompt reporting of Illness Symptoms and Side Effects (n=173) (facilitators for reporting side effects)				
1. Being able to reach a doctor without a hassle.	18.5%	.15	.21	.50
2. Having a doctor that is receptive.	12.4%	.59	.25	.0179

*p-value is significant, according to Benjamini-Hochberg procedure.

Table 5.7. Relationship of overall perceived barriers and facilitators to levels of the 8 self-management behavioral domains assessed by the VSMS.

		Perceived Difficulty			Number of Barriers Endorsed			Perceived Importance			Number of Facilitators Listed		
	n	*b*	*se*	*p*	*b*	*se*	*p*	*b*	*se*	*p*	*b*	*se*	*p*
Medication (self-administered)	187	-.09	.03	.0019*	-.13	.02	<.0001*	.33	.06	<.0001*	.04	.03	.17
Appointments with health professionals[1]	188	-.03	.04	.45	-.11	.03	.0013*	.12	.06	.07	.04	.05	.37
Tests and Immunizations[1]	197	-.11	.03	.0006*	-.14	.03	<.0001*	.35	.05	<.0001*	.03	.04	.49
Infection avoidance	139	-.21	.03	<.0001*	-.17	.04	<.0001*	.19	.06	.0039*	.09	.07	.22
Diet	151	-.18	.04	<.0001*	-.08	.03	.0093*	.21	.04	<.0001*	.10	.06	.10
Exercise	123	-.27	.05	<.0001*	-.15	.04	.0001*	.22	.06	.0005*	.24	.06	.0002*
Symptom monitoring	132	-.31	.05	<.0001*	-.05	.24	.84	.24	.06	.0003*	-.20	.22	.36
Adjusting activity level	182	-.14	.03	<.0001*	-.10	.03	.0002*	.13	.03	<.0001*	.07	.04	.11
Reporting symptoms[2]	172	-.24	.04	<.0001*	-.19	.03	<.0001*	.19	.04	<.0001*	.09	.07	.19
Reporting side	173	-.25	.04	<.0001*	-.17	.03	<.0001*	.21	.04	<.0001*	.10	.07	.20

effects[2]

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[1]Dependent variable was adherence to recommended health services.
[2]Dependent variable was prompt reporting of illness symptoms and side effects.
*p-value is significant, according to Benjamini-Hochberg procedure.

Table 5.8. Beta coefficients for the relationship of specific barriers to levels of eight self-management behaviors that became significant after deleting outliers.

	b	*se*	*p*
Adherence to Recommended Health Services (barriers for attending appointments)			
3. I had to travel a long way to get there. *(n=182)*	-.30	.10	0.002*
Adherence to Recommended Health Services (barriers for obtaining tests and immunizations)			
1. I had trouble scheduling the appointment at a time that was convenient for me. *(n=185)*	-.35	.10	<.0001*
Adherence to Infection Avoidance Recommendations			
1. My desire to participate in social/leisure activities got in the way. *(n=129)*	-.32	.12	0.011*
2. It was impossible for me to avoid people who were sick. *(n=127)*	-.33	.13	0.015*
5. Family responsibilities got in the way. *(n=130)*	-.38	.16	0.016*
Prompt reporting of Illness Symptoms and Side Effects (barriers to reporting symptoms)			
5. I was afraid of looking like a hypochondriac. *(n=163)*	-.51	.21	0.015*

*p-value is significant, according to Benjamini-Hochberg procedure.

CHAPTER SIX: SPOUSAL INFLUENCES ON ILLNESS SELF-MANAGEMENT IN

ADULTS LIVING WITH ANCA-ASSOCIATED SMALL VESSEL VASCULITIS:

THE ROLE OF ACCOMMODATIVE BEHAVIOR AND PERCEIVED SUPPORT

Abstract

Objective: The purpose of this investigation was to explore spouses' accommodative behavior as a supportive process that facilitates patients' chronic illness self-management behavior. Specifically, we explored whether patients' perceptions of general spouse support and/or illness-specific support mediated the relationship between spousal accommodative behavior and illness self-management behavior among adults living with ANCA-associated small vessel vasculitis (ANCA-SVV), a chronic autoimmune disease.

Methods: A total of 159 married adults living with ANCA-SVV completed a self-administered questionnaire that included measures of eight illness-specific self-management behaviors, their spouse's constructive accommodative and retaliation behavior, their own accommodative behavior, perceived spousal support (both general and illness-specific), and a variety of socio-demographic, clinical, and health variables. After examining descriptive statistics and bivariable correlations, path analysis was used to examine associations among spouse accommodative behavior, perceived spousal support, and patients' self-management behavior. Both general spouse support and illness-specific spouse support were examined as mediators of the effect of spousal accommodative behavior on patients' illness self-management behavior using Sobel's test for indirect effects. Alternative models, which allowed for direct effects of spousal accommodative behavior on self-management behavior in addition to indirect effects through perceptions of spouse support, were also assessed.

Results: Models allowing for direct effects of spousal accommodative behavior on self-management behavior, in addition to indirect effects through perceived spousal support, fit the data better compared to models allowing only indirect effects. In models investigating general spouse support as a mediator, spousal constructive accommodation was positively

associated with perceived support, and perceived support was, in turn, positively associated with medication adherence, adherence to recommended health services, and appropriate adjusting of activities in response to fatigue and illness symptoms. The indirect effects of spousal constructive accommodation on these three self-management behaviors via perceived general spouse support approached but did not reach statistical significance. In models investigating illness-specific spouse support as a mediator, spouse retaliation was negatively associated with perceived support, which was, in turn, positively associated with medication adherence, infection avoidance adherence, and appropriate adjusting of activities in response to fatigue and illness symptoms. Formal tests of these indirect effects again were not statistically significant.

Conclusions: Our results suggest that spousal accommodative behavior may serve as a relationship process by which perceptions of spousal support, both general and illness-specific, are determined. More research specifically investigating spouse support as a mediator of the effect of spousal accommodative behavior on patients' chronic illness self-management is warranted.

Introduction

Chronic illness self-management is defined as the daily tasks an individual performs, with the collaboration and guidance of health care providers, to control or reduce the impact of disease on his/her physical health status (N.M. Clark *et al.*, 1991). Self-management involves mastery of three types of activities: performing activities to manage the condition, making informed decisions about care, and applying skills to maintain adequate psychosocial functioning (N.M. Clark et al., 1991). Although one major component of illness self-management is treatment adherence, or "the extent to which a person's behavior – taking medication, following a diet, and/or executing lifestyle changes – corresponds with agreed recommendations from a health care provider" (WHO, 2003), self-management can also include performance of self-care behaviors not specifically recommended by health professionals, such as reducing stress in one's life, getting enough rest, and effectively communicating with physicians as needed. Specific self-management behaviors required by patients vary across conditions (N.M. Clark et al., 1991).

Performance of illness self-management behaviors are widely accepted as an important determinant of health outcomes in chronically ill individuals. A recent report by the World Health Organization stated that adherence to recommended health behaviors in chronic illness "…is a primary determinant of the effectiveness of treatment, because poor adherence attenuates optimal clinical benefit" (WHO, 2003, pp. 20). A recent meta-analysis of the relationship between treatment adherence and health outcomes revealed that good adherence reduced the risk of poor treatment outcome by 26% across all illnesses studied, and that treatment adherence was more strongly associated with positive outcomes in chronic versus acute illnesses (DiMatteo *et al.*, 2002).

Given the powerful impact of self-management behavior on health outcomes, it is not surprising that a large body of research has sought to understand the determinants of self-management behavior. This research has identified a number of patient-related, therapy-related, condition-related, health care team/health system, social, and economic factors that likely influence self-management behavior across a wide variety of illnesses (WHO, 2003). Among these factors, support from social network members has emerged as a consistent predictor of self-management behavior. Researchers have repeatedly found that patients' perceptions of social support provided by close others in their lives acts as a facilitator of treatment adherence in patients living with a variety of chronic illnesses (DiMatteo, 2004; Gallant, 2003; WHO, 2003). In this body of research, social support has typically been defined as a resource that individuals perceive to be available or provided in close relationships (Cohen *et al.*, 2000). While perceptions of general support and illness-specific support have both been linked to improved self-management behavior, a recent review of this research suggests that illness-specific support may be more strongly linked to health behavior than general social support (Gallant, 2003).

Many researchers have also argued that close romantic partners such as spouses play a key role in determining overall perceptions of available support (Reis & Collins, 2000; B.R. Sarason *et al.*, 1997) and enhancing behavioral and psychological outcomes among chronically ill patients (Revenson, 1993). Research evidence reinforces this argument. Not only are married individuals more likely to adhere to behavioral recommendations (DiMatteo, 2004), but both quantitative (Christensen *et al.*, 1992; Courneya *et al.*, 2001; Fraser *et al.*, 2003; Glasgow & Toobert, 1988; Wen *et al.*, 2004) and qualitative (El-Kebbi *et al.*, 1996; Maclean, 1991; Trief *et al.*, 2003) studies have implicated patients' perceptions of support

from family members and the spouse in particular as having an effect on patients' ability to carry out self-management behaviors. It is also important to note that negative interactions with spouses have been implicated as a detrimental influence on illness self-management behavior (N. M. Clark *et al.*, 1994; N. M. Clark & Nothwehr, 1997; Handron & Leggett-Frazier, 1994; Maclean, 1991), although this research has been primarily limited to qualitative studies.

Unfortunately, these consistent observations about the beneficial effect of naturally-occurring social support on treatment adherence have not translated into clear knowledge about how to intervene to improve support in close relationships, and, in turn, improve outcomes in chronic illness. Spousal support interventions specifically targeted at improving patients' self-management behavior are few in number. Related interventions designed to mobilize the support of key network members (often spouses) in the hopes of changing health behaviors and improving psychological adjustment to chronic illness have yielded modest and mixed results (Black *et al.*, 1990; Cutrona & Cole, 2000; Lichtenstein *et al.*, 1986; Park *et al.*, 2004; Riemsma *et al.*, 2003). One reason often cited for mixed intervention results is the simplistic treatment of social support as a perceived resource and a resulting lack of understanding about the mechanisms underlying its provision and receipt (Lassner, 1991).

In response to these findings, a new approach to studying social support and health has begun to emerge from the field of relationship science. This approach conceptualizes support as a dynamic *process* resulting from the motivations and skills of the provider and receiver of support as well as specific interactions in close relationships, rather than simply a *resource* (DeVellis *et al.*, 2003). Such an approach is potentially more informative with regard to

intervention development because it may offer more specific guidance on how to favorably change patients' perceptions of spousal support, which has proven to be very difficult in many couples-focused interventions (Lichtenstein et al., 1986). Two constructs from this approach – transformation of motivation and accommodative behavior – are a promising alternative to understanding how support may affect patients' illness self-management (Lewis et al., in press).

The construct of transformation of motivation comes from Interdependence Theory (Rusbult & VanLange, 1996), which is commonly used to guide research on outcomes in close relationships (e.g., relationship maintenance or dissolution) (DeVellis et al., 2003). Interdependence Theory suggests that spouses experience individual outcomes as well as shared outcomes related to the goals of their relationship, and that spouses mutually influence each others' outcomes. A central concept in Interdependence Theory is transformation of motivation (Yovetich & Rusbult, 1994), or when an "individual internalizes social interactions from another person, and thus acts in pro-relationship, health-enhancing ways rather than acting in a purely self-interested manner" (DeVellis et al., 2003). When transformation of motivation has occurred, each partner's goals become the other partner's goals, and each partner's behavior reflects these mutual goals.

One commonly used indicator of whether transformation of motivation has occurred is spouses' accommodative behavior, which refers to the extent partners react constructively rather than destructively when the other acts in a relationship-threatening way (i.e., with hostility or criticism) (Rusbult *et al.*, 1991). Accommodation is a multi-dimensional construct, in that individuals' responses to destructive acts by their partners can be constructive or destructive in terms of their effects on the relationship, as well as active or

passive with regard to their effects on the problem at hand (see Figure 1). Rusbult and

colleagues (Rusbult & Verette, 1991) have defined four patterns of accommodative behavior:

voice, loyalty, exit, and neglect. Voice is characterized by active, constructive attempts to

resolve the situation (e.g., discussing problems and suggesting solutions). Loyalty is

characterized by a passive, constructive response in which the individual passively expects

and waits for things to improve (e.g., simply waiting until the partner's mood and behavior

improves). Exit is an active, destructive response (e.g., yelling, threatening to leave, physical

abuse), while neglect is a passive, destructive response (e.g., ignoring the partner, criticizing

the partner for issues unrelated to the apparent conflict). Voice and loyalty are together

considered to be "constructive accommodation" while exit and neglect are considered to be

"retaliation" (Rusbult et al., 1998).

Although accommodation has not previously been studied in the context of chronic illness,

several studies have been conducted to specify the impact of accommodative behavior on

couples' adjustment, distress, and functioning. Like a lot of social psychological studies,

these studies have primarily been conducted with healthy young couples in dating

relationships or in the early years of marriage and therefore may not have produced results

that are generalizable to older couples who have been married for many years or who are

facing a long-term illness. However, this body of research has shown good evidence that

higher levels of constructive accommodation and lower levels of retaliation on behalf of *both*

spouses are associated with higher levels of couple functioning, including greater trust and

dyadic adjustment (Rusbult et al., 1986; Rusbult et al., 1991; Wieselquist et al., 1999). If

accommodative behavior on behalf of both spouses jointly determine dyadic adjustment, then

it is not unreasonable to hypothesize that accommodation in marital relationships may also

affect individuals' perceptions of support from partners. Furthermore, if accommodation is a

mechanism underlying perceptions of spouse support, then outcomes specific to the context

of chronic illness, such as patients' self-management behavior, could also be affected by

accommodation. There is also evidence from qualitative studies suggesting that interactions

with spouses that patients perceive as negative may act as a barrier to performing self-

management behaviors (Clark & Nothwehr, 1997; Clark, et al., 1994; Maclean, 1991;

Handron & Leggett-Frazier, 1994). This further suggests that spousal accommodative

behavior may be relevant to patients' self-management behavior, either directly or via an

effect on perceived spouse support. Thus, the primary purpose of this study is to investigate

spouses' accommodative behavior as part of a supportive process that facilitates patients'

chronic illness self-management behavior.

Conceptual Model

Figure 2 depicts a conceptual model of how accommodation in marital relationships is

hypothesized to affect patients' self-management behavior. The model combines ideas from

the traditional "direct effects" model of the influence of social support on health behavior, in

which perceived support directly influences health behavior (Burman & Margolin, 1992),

and research drawing on Interdependence Theory, in which accommodative behavior on

behalf of both spouses influences perceptions of one's relationship functioning. Considering

the model from left to right, spouses' and patients' accommodative behavior are

hypothesized to be correlated with one another (as depicted by the double-headed curved

arrows). Paths are then drawn leading from spouse retaliation and constructive

accommodation and patient retaliation and constructive accommodation to patients'

perceptions of spousal support. This reflects the hypothesis that perceptions of spousal

support are jointly determined by each spouse's accommodative behavior. Finally, patient

perceptions of spousal support are then shown to directly influence patients' treatment

adherence. Thus, the conceptual model suggests four hypotheses that will be explicitly tested

in this study:

Hypothesis 1: Higher levels of retaliation by spouses will be negatively associated with

patients' perceived spousal support, independent of the spouse's level of constructive

accommodation and the patient's own accommodative behavior.

Hypothesis 2: Higher levels of constructive accommodation by spouses will be positively

associated with patients' perceived spousal support, independent of the spouse's level of

retaliation and the patient's own accommodative behavior.

Hypothesis 3: Patients' perceived spousal support will be positively associated with

patients' self-management behavior.

Hypothesis 4: Patients' perceived spousal support will completely mediate the relationship

between spousal accommodative behavior and patients' self-management behavior.

Because prior research suggests that perceived general spouse support and perceived

illness-specific support may be differentially related to the patient's self-management

behavior, both types of support were investigated as mediators of the relationship between

spousal accommodative behavior and patient's self-management behavior.

Alternative Model

It should be noted that the lack of arrows leading directly from the accommodation

variables to self-management behavior in Figure 2 implies that accommodation in marital

relationships exerts its effect on self-management entirely through patient perceptions of

spousal support. It is possible that spousal accommodative behavior directly influences

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patients' self-management or operates through pathways other than social support. Therefore, an alternative model, shown in Figure 3, including direct paths from spousal accommodative behavior to patient self-management behavior were also evaluated in this study.

To formally evaluate these conceptual models, we studied adults living with ANCA-associated small vessel vasculitis (ANCA-SVV), a group of rare autoimmune conditions characterized by inflammation and necrosis of blood vessels primarily in the respiratory tract and kidneys (Watts & Scott, 1997. ANCA-SVV is a potentially fatal, relapsing illness, with an often progressive but unpredictable course (Koutantji *et al.*, 2000). There is no cure for the illness, but treatment with immunosuppressive medications induces remission in 80-100% of patients (Langford, 2003). Patients may experience full remission, partial remission, and/or minor or major relapses (Jayne, 2003), although the risk of relapse and morbidity from the disease and its ongoing, highly toxic treatment remains high even in remission (Hoffman *et al.*, 1998). Treatment regimens for ANCA-SVV are long-term, complex, frequently altered by physicians to address changes in disease activity, and associated with serious side effects. Together with their physicians, patients typically agree to follow a demanding regimen which includes a variety of self-care tasks, including medication-taking, obtaining vaccinations, regular use of health services and communication with their physicians, and a range of other lifestyle changes. These regimens are often adjusted based on current disease activity and the occurrence of side effects and complications. While ANCA-SVV is a rare condition, the behaviors involved in its management are the same behaviors required of other common chronic illnesses, thus making it an appropriate illness context for examining spousal influences on self-management behavior.

Methods

Design

This investigation was conducted as part of the Vasculitis Self-Management Study, which was a cross-sectional, observational study of adults living with ANCA-associated small vessel vasculitis (ANCA-SVV). All participants completed a self-administered, mailed questionnaire assessing various aspects of ANCA-SVV self-management, psychosocial issues, and socio-demographics. Participants were provided with a $10 gift (in the form of cash or a gift card) after completing and returning the questionnaire.

Sample

To be eligible for the Vasculitis Self-Management Study, patients were required to be 18 years or older, be able to read and write in English, and have received a diagnosis of a form of ANCA-SVV or a closely related condition. In addition, the sample for these analyses was limited to individuals who reported being currently married. Adult patients meeting eligibility criteria were recruited for participation by posting announcements about the study in a variety of places. First, patients 18 years or older who were part of an ANCA-SVV and glomerulonephritis patient disease registry at the lead author's institution and had signed long-term consent to be contacted about vasculitis research were invited to participate via letter. Announcements about the study were also placed in newsletters targeted toward ANCA-SVV patients and on several ANCA-SVV-related websites. Finally, ANCA-SVV patients who had volunteered for another study in which the lead author was involved were asked if they were interested in hearing about other ANCA-SVV research, and if they responded positively, they were invited to participate in this study. A total of 305 interested patients were identified through these methods and mailed study consent forms. Of these,

240 (79%) signed and returned the consent form, and 218 (71%) completed and returned the study questionnaire. Two participants were subsequently excluded because they or their doctor reported a primary diagnosis of lupus and one participant was excluded because he reported an unknown diagnosis which his physician later indicated was not vasculitis-related. Of the 215 eligible patients returning completed questionnaires, 159 (74%) reported being currently married and thus comprised the final study sample.

<p align="center">*Measures*</p>

Vasculitis Self-management

ANCA-SVV self-management was measured using the Vasculitis Self-Management Scale (VSMS). The initial testing of the VSMS was conducted using the current study's sample and is described in detail, along with pilot work for developing the VSMS, elsewhere (Thorpe, et al., in preparation). The VSMS was designed to assess individuals' treatment adherence (i.e., performance of behaviors directly recommended by health professionals to them for controlling their condition), and also their performance of self-care behaviors generally recommended for all ANCA-SVV patients. The VSMS includes six adherence subscales and two self-care subscales that assess individuals' self-reported performance of illness self-management behaviors found to be relevant for the majority of individuals living with ANCA-SVV: 1) self-administered medication adherence, 2) adherence to recommended health services, 3) adherence to infection avoidance recommendations, 4) dietary adherence, 5) exercise adherence, 6) adherence to symptom monitoring recommendations, 7) adjusting activities in response to fatigue or symptoms, and 8) prompt reporting of new or increased illness symptoms or side effects to a health professional. If a specific self-management behavior was not relevant for a respondent (i.e., because the behavior had not been

recommended by a physician or the patient did not have any symptoms during the recall period), he/she was instructed to skip the section. Two different recall periods were used in the VSMS, depending on the frequency with which patients are typically required to perform the behavior. For behaviors that are typically performed on a daily basis (e.g., medication-taking, following dietary guidelines), patients were asked to report the frequency with which they performed the behavior according to their physicians' recommendations in the past four weeks. This recall period was thought to be long enough to produce sufficient opportunity for non-adherence and in turn, variation in responses among respondents, but short enough to produce reliable and accurate self-reports of behavior. For behaviors that are performed with varying frequency and/or much less often than daily (e.g., attending appointments) patients were asked to report their behavior during the past year. The response format varies across items in the VSMS, but consistently uses five-point Likert scales (e.g., *none of the time* to *all of the time*). Several items pertaining to each adherence behavior are negatively worded and reverse-scored, so that ultimately, higher scores on each item correspond to higher levels of adherence. Internal consistency reliability (i.e., Cronbach's alpha) for the eight self-management subscales ranged from acceptable (0.62) to excellent (0.95), as shown in the bottom part of Table 6.1.

Spousal Accommodative Behavior

Patients' perceptions of their spouses' accommodative behavior were assessed using a slightly modified version of the My Partner's Responses to Relationship Problems scale (unpublished scale by Rusbult, available at www.unc.edu/depts/socpsych/cr/Accommodation.html). This scale measures an individual's perceptions about his/her spouse's responses to the individual's own destructive (exit or

neglect) behavior. That is, the patient was asked to report how the healthy spouse tends to respond to the patient's exit or neglect behavior. Two subscale scores were created to summarize spousal accommodative behavior. The constructive accommodation subscale consisted of average scores for eight items describing voice and loyalty accommodative behavior, and the retaliation scale consisted of average scores for eight items describing exit and neglect behavior. Because the original My Responses to Relationship Problems scale was designed for use with couples who may be unmarried (i.e., dating) or newlyweds, we adapted the scale to be more appropriate for couples who have been married for many years. References to breaking up or ending the relationship (an exit response) were altered to account for the fact that couples who have been married for many years would likely endorse this response only very rarely, compared to dating couples. For example, "my partner considers breaking up" was changed to "my partner thinks his/her life would be better without me." These altered items are still thought to tap into exit responses, but should be more appropriate for couples who have been married for many years. Finally, we also adapted the stems of each item to pertain to accommodative behavior in the specific context of chronic illness (e.g., the original item "when I'm upset and say something mean, my partner tries to patch things up and solve the problem" was altered to state "when I am not feeling well and say something mean, my spouse tries to patch things up to solve the problem").

The perceived partner constructive accommodation and retaliation scores have been used most often in previous research, with good evidence of reliability (Rusbult et al., 1998; Rusbult et al., 1991; Wieselquist *et al.*, 1999). Cronbach's alphas for the spousal constructive accommodation and retaliation scales in this study were 0.88 and 0.84,

respectively. In addition, these two subscales have demonstrated validity in previous studies. For example, correlations between own and partner reports for constructive accommodation (rs = 0.35, 0.51, 0.42) and retaliation scales (rs = 0.57, 0.52, 0.45) were also demonstrated to be high across three time points in one study (Rusbult et al., 1998), and own and partner reports of accommodative behavior similarly and significantly predicted relationship outcomes such as trust and dyadic adjustment in another study (Wieselquist et al., 1999).

Patient's Accommodative Behavior

Patients' perceptions of their own accommodative behavior were assessed using the My Responses to Relationship Problems scale (unpublished scale by Rusbult, available at www.unc.edu/depts/socpsych/cr/Accommodation.html). This scale is exactly parallel to the My Partner's Responses to Relationship Problems scale but asks individuals to report on their *own* responses to their partners' exit or neglect behavior. That is, the patient reports his/her own accommodative behavior. The scoring methods are the same as for the partner scale described above, and the wording of specific items was also revised in the ways described above. The own constructive accommodation and retaliation subscales have also demonstrated adequate internal consistency reliability in previous research (Rusbult et al., 1998; Rusbult et al., 1991; Wieselquist et al., 1999). Cronbach's alphas in this study were 0.91 and 0.83 for the own constructive accommodation and retaliation subscales, respectively. In addition to the evidence for validity described above, the own accommodation subscales have also demonstrated good criterion-related validity, based on substantial correlations with observers' ratings of accommodative behavior in videotaped interactions, accommodation in matrix games, and accommodation in a moral dilemma task (Rusbult et al., 1991).

Perceptions of General Spouse Support

Patients' perceptions of the availability of general support from their spouses were measured using the spouse-specific version of the Social Provisions Scale (Cutrona & Russell, 1987). This measure is designed to assess six supportive functions provided by social network members: reliable alliance (belief that others can be relied on for tangible assistance), attachment (emotional closeness and a sense of security provided by others), guidance (advice or information available from others), nurturance (the sense that others rely on the patient for their well-being), social integration (a sense of shared interests and concerns with others), and reassurance of worth (others' recognition of the patient's competence, skills, and value). Previous research has shown higher summed scores on the Social Provisions Scale to be predictive of better treatment adherence among end-stage renal disease patients (Moran *et al.*, 1997) and higher scores on the attachment subscale to be predictive of improved medication adherence among individuals with HIV (Catz *et al.*, 2000). Because the overall scale shows considerably higher reliability than the individual subscales, Wills and Shinar (2002) suggest that the overall scale score may be a more appropriate measure of overall perceived support than the individual subscales. In addition, prior research with end-stage renal disease patients has shown the overall summary score to be positively associated with treatment adherence (Moran, et al., 1997). Therefore, the overall score on the Social Provisions Scale is used in these analyses (Cronbach's $\acute{\alpha} = 0.89$).

Perceptions of Illness-specific Spouse Support

An adapted version of the spouse-specific Social Provisions Scale was used to assess patients' perceptions of available spouse support specific to ANCA-SVV self-management. Items from the Social Provisions Scale have been adapted, with good results, to measure

health-related, behavior-specific support in other studies (Darbes & Lewis, in press).

Because instrumental and emotional support are most relevant to self-management and items

measuring other types of support were not easily adapted to become illness-specific items,

only items from the reliable alliance, guidance, and attachment subscales of the Social

Provisions Scale were adapted for this measure. Responses to all 12 items were averaged,

after appropriate reverse-scoring, to create a summary score (Cronbach's ά was 0.94). A

principal components analysis supported the uni-dimensionality of this adapted scale, in that

a one-factor solution accounted for 65% of the total variance and all items had loadings of at

least .64 on a single factor (complete results available upon request).

Socio-demographics

In addition to completing the VSMS, patients were asked to provide basic socio-

demographic information, including gender, race/ethnicity, age (in years), education level (in

years), and duration of their current marriage (in years).

Clinical and Health Variables

A number of clinical and health variables were also assessed to further characterize the

patient sample. In addition to their specific ANCA-SVV-related diagnosis (e.g., Wegener's

granulomatosis, microscopic polyangiitis), patients were asked to report their history, if any,

of dialysis and kidney transplant. Illness duration, in months, was assessed by asking

participants to report the month and year of diagnosis with ANCA-SVV, and then subtracting

these values from the month and year of questionnaire completion. Current disease activity

was assessed by physician report. Physicians were asked to rate on a scale from 1 (not active

at all/remission) to 10 (extremely active) how active the patient's vasculitis currently was.

Morbidity and damage from vasculitis were also assessed via physician report using a similar 10-point scale (1=no damage to 10=extreme damage). Complexity of the patient's treatment regimen was measured using the total number of self-management behaviors the patient reported as having been recommended by a physician. Current health status was also assessed using the General Health Scale of the SF-36, Version 2.0, scored using the RAND method (J. E. Ware, Jr. & Sherbourne, 1992) (Cronbach's $\acute{\alpha}$= .82).

Social Desirability Bias

Social desirability, or the desire of respondents to appear in ways that society regards as positive, was assessed in the study questionnaire using the short form of the Marlowe-Crowne Social Desirability Scale (Strahan & Gerbasi, 1972). This scale is widely used in psychometric studies to determine whether answers to self-reported questionnaires are influenced by respondents' desires to appear in socially desired ways. Cronbach's $\acute{\alpha}$ in this study was 0.77.

Analysis

Analytic Approach

To characterize the sample, descriptive statistics for all variables described above were first examined. For categorical variables, descriptive statistics included the overall *n* and *%* in each category. For continuous variables, *M, SD*, skewness, and kurtosis were examined. Bivariable (pairwise) correlations among all study variables were then examined. All univariable and bivariable analyses were conducted using SAS Version 9.13 (SAS Institute, Inc., Cary, NC).

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We then used MPLUS software (version 3.12, Muthén and Muthén, 2005) to conduct path analysis to evaluate the hypotheses implied by the conceptual models described above. Path analysis is a type of structural equation modeling that involves estimating a system of simultaneous regression equations, with one equation per endogenous (dependent), measured variable. A major advantage of path analysis is that it can be used to produce estimates of each path depicted in a conceptual model, including the total effects, direct effects, and indirect effects of independent variables on dependent variables. In order for statistical tests of estimates produced by path analysis to be valid, observations must be independent of one another and data should adhere to a multivariate normal distribution (Hoyle, 1995). Because a few variables in the models demonstrated significant non-normality (i.e., skewness > 2 and kurtosis > 7; (Curran *et al.*, 1996)), thus suggesting that the data were likely not multivariately normally distributed, we used maximum likelihood estimation with robust standard errors in all path models.

Handling of Missing Data in the Path Analyses. It was anticipated that all items would have some proportion of data missing, because not all self-management behaviors were expected to be relevant for all patients and as a result, patients would, as instructed, skip some items when completing the questionnaire. It is assumed that missing data due to the skip patterns are missing at random (MAR), and therefore, ignorable. MAR assumes that the probability that data for a given variable are missing does not depend on the value of the variable itself, after other observed variables are controlled, although there is no test to determine whether this is the case (Allison, 2003). That is, the probability that a particular value for a variable is missing may depend on the values of other variables in the model, but is assumed to be unrelated the value of variable that is missing. In this study, we assumed

that the probability of missing data on the self-care items is not related to the individual's level of self-care, but instead is directly the result of the behavior not being relevant to the individual's specific illness situation, or possibly other demographic, clinical, or psychosocial variables controlled in the path models, resulting in data that are MAR. When data are MAR and/or ignorable, a variety of techniques can be used when conducting path analysis: listwise deletion, pairwise deletion, condition mean imputation, multiple imputation (MI), and full-information maximum likelihood (FIML) (Allison, 2003; Muthen et al., 1987). Of these methods, FIML and MI are currently the preferred methods for handling missing data in structural equation modeling because of their improved accuracy in estimating variances and covariances (Allison, 2003). While both FIML and MI are available in MPLUS when maximum likelihood estimation is used, MI was used in this study because of its compatibility in MPLUS with our preferred statistical tests of model fit (i.e., the Satorra-Bentler scaled chi-square statistic, described below).

Statistical models. A total of four models were tested, including two models for each operationalization of social support (i.e., general perceived spousal support and illness-specific spouse support). To evaluate general perceptions of spousal support as a mediating variable, we estimated Model A-1 and Model A-2. In both of these models, we estimated nine simultaneous regression equations, one for each dependent variable in the model (the eight self-management variables plus general perceived spousal support). Patient socio-demographics (gender, white versus other race/ethnicity, age, and education level), marriage duration, disease duration, regimen complexity, social desirability bias, and physician ratings of the patient's current disease activity and damage were included in all nine regression equations as independent variables to control for their potential confounding effects on the

relationship between accommodative behavior and all nine of the dependent variables, as well as spousal support and the self-management dependent variables. In the first regression equation, perceived spousal support was the dependent variable, and the accommodation variables (i.e., spouse retaliation and constructive accommodation, patient retaliation and constructive accommodation) and control variables were independent variables. The other eight equations each included one of the eight self-management variables as dependent variables, and general perceived spousal support and the control variables as independent variables. In essence, Model A-1 posits that the effect of marital accommodation on patient's self-management behavior is fully mediated by the patient's perceived general spousal support. The regression equations are shown below. The subscript "p" refers to patient attributes; the subscript "s" refers to spouse attributes (as reported by the patient).

Model A-1, Equation 1:

$$\text{Support}_p = \beta_0 + \beta_1 \text{retaliation}_s + \beta_2 \text{constructive}_s + \beta_3 \text{retaliation}_p + \beta_4 \text{constructive}_p + \beta_5 \text{control}_p$$

Model A-1, Equations 2-9:

$$\text{Self-management behavior}_p = \beta_0 + \beta_1 \text{Support}_p + \beta_2 \text{control}_p$$

Model A-2 was then tested as an alternative to Model A-1. In Model A-2, two additional paths were estimated to allow for the possibility that spousal accommodative behavior (both constructive accommodation and retaliation) has direct effects on patient self-management, in addition to indirect effects through patient perceptions of spousal support. Model A-1 is thus "nested" within (and can therefore be directly compared with) Model A-2, meaning that Model A-2 includes all of the same pathways as Model A-1, plus two additional pathways

not included in Model A-1. That is, Equation 1 above remained unchanged in Model A-2, but Equations 2-9 included the addition of the two spousal accommodation variables:

Model A-2, Equations 2-9:

$$\text{Self-management behavior}_p = \beta_0 + \beta_1 \text{Support}_p + \beta_1 \text{retaliation}_s + \beta_2 \text{constructive}_s + \beta_3 \text{control}_p$$

To evaluate illness-specific spousal support as a mediating variable, we also estimated Models B-1 and B-2. These models are identical to Models A-1 and A-2, respectively, except that illness-specific spousal support was substituted for general perceived spousal support in all regression equations.

Assessment of model fit. Three fit indices, including two measures of absolute fit and one measure of incremental fit, were used to evaluate the omnibus fit of each model described above, in line with current recommendations for evaluating model fit (Curran *et al.*, 1996; Hu & Bentler, 1999) and reporting the results of structural equation modeling (Hoyle & Panter, 1995). First, we used the Satorra-Bentler scaled chi-square statistic to assess absolute fit, or the extent to which the observed covariances from the study data fit the covariances implied by the conceptual model (Hoyle & Panter, 1995). Higher values of the chi-square statistic (and lower corresponding p-values) indicate worse model fit. The Satorra-Bentler scale chi-square was used instead of the uncorrected chi-square because of the suspected multivariate non-normality. When data are multivariate non-normal, the chi-square statistic tends to be positively biased and results in over-rejection of the null hypothesis that the data fit the model well. The Satorra-Bentler chi-square adjusts for the extent of non-normality in the data and has been shown to perform well under a variety of conditions, including small sample sizes (Curran et al., 1996).

We also used one additional measure of absolute fit (the standardized root mean squared residual, or SRMR; (Bentler, 1995)) and one measure (the Comparative Fit Index, or CFI); of incremental fit, or the extent to which the proposed model outperforms the null, independence model (i.e., a model where there are no relationships between variables) when compared to the observed covariances (Hoyle & Panter, 1995). When sample size is less than 250, SEM experts (Hu & Bentler, 1999) recommend using SRMR in combination with one additional incremental fit index, such as the comparative fit index (CFI), to assess overall model fit. They suggest rejecting model fit when the SRMR $>.09$ and the CFI $< .95$, based on extensive simulation studies under various conditions of sample size, model misspecification, and multivariate normality (Hu & Bentler, 1999). Thus, these cut-offs were used in this study in conjunction with the Satorra-Bentler chi-square to determine overall model fit for the four models being tested.

In addition, the nested competing models were compared to one another using the chi-square difference test, in line with current recommendations (Hoyle & Panter, 1995). That is, Model A-1 was compared to Model A-2, and Model B-1 was compared to B-2, to determine if estimation of direct paths from spousal accommodative behavior to self-management improved model fit over models in which perceived support completely mediated the relationship between spousal accommodative behavior and self-management. In this test, the chi-square value for the more complex model (i.e., one with fewer degrees of freedom, such as Model A-2) is subtracted from the chi-square value for the less complex model (such as Model A-1), and this difference is evaluated against the chi-square distribution. In this study, chi-square difference values corresponding to a p-value of $<.05$ were chosen as the *a priori*

cut-off for significance, which would indicate preference for the more complex model with additional estimated direct paths from spousal accommodative behavior to self-management.

Assessment of parameter estimates. Once the final models were chosen, parameter estimates within each preferred model were examined to evaluate specific hypotheses. Critical ratios of greater than or equal to +/- 1.96, which correspond to a p-value of <.05, were used to evaluate the individual significance of parameter estimates for paths leading from accommodative behavior to perceived spousal support, and from perceived spousal support to the eight self-management behaviors.

Assessment of indirect effects/mediation. Patient perceptions of spousal support (both general and illness-specific) were specifically assessed as a mediator of the effect of the spousal accommodation variables on self-management behavior using Sobel's products of coefficients approach (Sobel, 1982). In this approach, the coefficient for the path leading from the independent variable to the mediator is multiplied by the coefficient for the path leading from the mediator to the dependent variable to produce an estimate of the mediation effect. This estimate is then divided by its standard error (as computed using Sobel's formula) and compared to a standard normal distribution. Although parameter estimates are usually available in MPLUS using Sobel's approach, they are not available when multiple imputation is used to handle missing data. Thus, we used an available online interactive tool for assessing significance of indirect effects using Sobel's approach (found at http://www.unc.edu/~preacher/sobel/sobel.htm).

<div align="center">Results</div>

<div align="center">*Univariable Statistics*</div>

Descriptive statistics for patient and spouse socio-demographic variables are shown in Table 6.1. Approximately half the sample (50.3%) was male, and a substantial majority (95.6%) reported being White/Caucasian. The mean age of the sample was 56.5 years (SD=12.8, range 29-83), and the mean level of education was 14.7 years (SD=2.3), or some college. The socio-demographic profile of spouses was very similar to that of patients, in that mean age was 57.7 years (SD=12.6), mean education level was 14.3 years (SD=2.4), and 92.5% of spouses were White/Caucasian. The majority of the patient sample (73.0%) reported a diagnosis of Wegener's granulomatosis, and the mean duration of living with ANCA-SVV was 77.1 months (SD=72.5), or just over six years (range=3 months to 41.6 years). The average score on the General Health scale of the SF-36 was 46.8 (SD=23.4), which is substantially lower than the mean of 71.9 reported for the general U.S. population (J.E. Ware, Jr., 2000).

Descriptive statistics for psychosocial characteristics (including perceived spousal support), accommodative behavior, and self-management behaviors are shown in the bottom portion of Table 6.1. In general, patients had positive perceptions of the available support from their spouses, both general (M=129.8, SD=14.3) and illness-specific (M=5.6, SD=0.7). With regard to their own and their spouse's behavior, patients reported that constructive accommodation occurred quite frequently and retaliation occurred relatively infrequently. Self-reported performance of self-management behaviors was highest for adherence to recommended health services (M= 4.6, SD=0.5) and medication adherence (M=4.5, SD=0.5), and lowest for exercise adherence (M=3.1, SD=1.0) and prompt reporting of illness symptoms and side effects (M=3.2, SD=1.1).

Three study variables, including two dependent variables, displayed significant skewness and kurtosis: time since diagnosis (skewness = 2.22, kurtosis = 7.81), illness-specific perceived spousal support (skewness = -3.29, kurtosis = 14.38) and adherence to recommended health services (skewness = -2.30, kurtosis = 7.15). These results led to our decision to use maximum likelihood estimation with robust standard errors and the Satorra-Bentler scaled chi-square statistic for assessing omnibus model fit, as described above.

Bivariable Correlations

Table 6.2 shows the pairwise correlations for all study variables included in the path analysis regression models. Patient ratings of spouse constructive accommodation and retaliation as well as ratings of their own constructive accommodation and retaliation were all moderately to strongly correlated with one another (i.e., the absolute value of the *r*'s ranged from 0.27 to 0.75, all *p*'s <.001). Patients' general perceived spouse support was positively correlated with spouse constructive accommodation (*r*=0.46, *p*<.001) and their own constructive accommodation (*r*=0.27, *p*<.001), and negatively correlated with spouse retaliation (*r*=-0.39, *p*<.001) and their own retaliation (*r*=-0.54, *p*<.001). The correlations between illness-specific spouse support and spouse accommodative behavior followed a similar pattern, in that illness-specific spouse support was positively correlated with spouse constructive accommodation (*r*=0.48, *p*<.001) and one's own constructive accommodation (*r*=0.24, *p*<.001), and negatively correlated with spouse retaliation (*r*=-0.52, *p*<.001) and one's own retaliation (*r*=-0.46, *p*<.001). While both general (*r*=0.22, *p*<.01) and illness-specific support (*r*=0.32, *p*<.001) were positively correlated with medication adherence, and illness-specific support was also positively correlated with infection avoidance adherence (*r*=0.21, *p*<.05), support was not significantly associated with the majority of the self-

management variables in the bivariable analyses. Spouse accommodative behavior was also

not significantly associated with the self-management variables, but patient ratings of their

own retaliation were significantly and negatively associated with infection avoidance

adherence (r=-0.21, p<.05), symptom monitoring adherence (r=-0.23, p<.05), and reporting

symptoms and side effects (r=-0.18, p<.05).

Assessment of Model Fit

Table 6.3 shows the omnibus model fit indices for all four models tested in this study,

along with the results of the chi-square difference tests comparing nested models to one

another.

The first row of Table 6.3 shows results for Models A-1 and A-2. Regarding Model A-1,

both the Satorra-Bentler chi-square (χ^2 (32, 159) = 71.386, p<.001); and the CFI (CFI=0.90)

suggested poor model fit, although the SRMR (SRMR=0.03) was acceptable. The chi-square

difference test for Model A was significant (χ^2 (16, 159) = 39.676, p<.001), suggesting that

Model A-2 fit the data better than Model A-1. The omnibus model fit indices for Model A-2

also suggested better model fit. Although the Satorra-Bentler chi-square statistic remained

significant in Model A-2 (χ^2 (16, 159) = 31.710, p<.05), the p-value was reduced and the CFI

suggested good model fit (CFI=0.96) along with the SRMR (SRMR=0.02). Thus, Model A-

2 was accepted as the preferred model for examining general spousal support as a mediator

of the effect of spousal accommodative behavior on self-management.

The second row of Table 6.3 shows results for Models B-1 and B-2. Regarding Model B-1,

both the Satorra-Bentler chi-square (χ^2 (32, 159) = 65.352, p<.001) and the CFI (CFI=0.91)

indicated poor model fit, although the SRMR was once again acceptable (SRMR=0.03). The

chi-square difference test for Model B was significant (χ^2 (16, 159) = 29.66, p<.05),

suggesting that Model B-2 fit the data better than Model B-1. As with Model A, the omnibus model fit indices for Model A-2 also indicated better model fit. While the Satorra-Bentler chi-square test statistic remained significant in Model B-2 (χ^2 (16, 159) = 35.692, $p<.01$), the p-value was smaller than in Model B-1 and the CFI suggested good model fit (CFI=0.95) along with the SRMR (SRMR=0.02). Thus, Model B-2 was accepted as the preferred model for examining illness-specific support as a mediator of the effect of spousal accommodative behavior on self-management.

<p style="text-align:center">Assessment of Parameter Estimates</p>

Model A-2

The individual parameter estimates, including unstandardized beta coefficients and standard errors, for each path estimated in Model A-2 are shown in Table 6.4. Each column represents estimates for one of the nine simultaneous regression equations (one for each endogenous variable) estimated in the path analysis.

The parameter estimates in the first *column* of Table 6.4 represent the effects of the accommodation and control variables on patients' perceived general spouse support. As expected, spouse constructive accommodation was positively associated with perceived general spouse support (*b*=3.36, *se*=1.26, *p*<.01). Contrary to expectations, spouse retaliation was not significantly associated with perceived general spouse support (*b*=-0.55, *se*=1.58), but higher levels of the patient's own self-reported retaliation significantly predicted lower levels of perceived general spouse support (*b*=-4.82, *se*=1.26, *p*<.01).

The parameter estimates in the first *row* of Table 6.4 represent the effect of perceived general spouse support on the eight self-management behaviors. Significant effects were found for three self-management behaviors. As expected, perceived general spouse support

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was positively associated with medication adherence ($b=0.007$, $se=0.003$, $p<.05$), adherence to recommended health services ($b=0.006$, $se=0.003$, $p<.05$), and adjusting activities in response to fatigue or symptoms ($b=0.010$, $se=0.004$, $p<.01$). Contrary to expectations, perceived general spouse support did not significantly predict adherence with regard to infection avoidance, diet, exercise, or symptom monitoring, or prompt reporting of illness symptoms and side effects to a health professional.

The parameter estimates in the second and third rows of Table 6.4 represent the direct effects of spouse retaliation and constructive accommodation on self-management behavior. While the results of the model comparison tests described above recommended allowing these paths to be estimated, none of these paths were statistically significant, with one exception. Spouse retaliation had a significant, *positive* direct effect on exercise adherence; that is, higher levels of spouse retaliation predicted higher levels of exercise adherence.

Model B-2

The individual parameter estimates, including unstandardized beta coefficients and standard errors, for each path estimated in Model B-2 are shown in Table 6.5. Each column represents estimates for one of the nine simultaneous regression equations (one for each endogenous variable) estimated in the path analysis.

The parameter estimates in the first *column* of Table 6.5 represent the effects of the accommodation and control variables on patients' perceived illness-specific spouse support. As expected, spouse retaliation was negatively associated with illness-specific spouse support ($b=-0.180$, $se=0.090$, $p<.05$). Contrary to expectations, spouse constructive accommodation was not significantly associated with illness-specific spouse support

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(b=0.109, se=0.068). Patients' reports of their own accommodative behavior were not significantly related to perceived illness-specific support.

The parameter estimates in the first *row* of Table 6.5 represent the effect of perceived illness-specific spouse support on the eight self-management behaviors. Significant effects were found for three self-management behaviors. As expected, perceived illness-specific spouse support was positively associated with medication adherence (b=0.232, se=0.064, p<.01), infection avoidance adherence (b=0.277, se=0.139, p<.05), and adjusting activities in response to fatigue or symptoms (b=0.195, se=0.085, p<.05). Contrary to expectations, illness-specific spouse support did not significantly predict adherence with regard to recommended health services, diet, exercise, or symptom monitoring, or prompt reporting of illness symptoms and side effects to a health professional, although its relationship with dietary adherence was marginally significant in the expected direction (b=.180, se=.108, p<.10).

The parameter estimates in the second and third rows of Table 6.5 represent the direct effects of spouse retaliation and constructive accommodation on self-management behavior. While the results of the model comparison tests described above recommended allowing these paths to be estimated, none of these paths were statistically significant.

Formal Assessment of Indirect Effects

The test statistics produced by the Sobel tests for indirect effects of spousal accommodative behavior on self-management behavior, via perceived social support, are shown in Table 6.6. No significant indirect effects were identified using Sobel's approach, although three indirect paths were marginally significant at p<.10. In Model A-2, the indirect effect of spouse constructive accommodation on medication adherence via perceived general

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spouse support approached statistical significance (t=1.756, p=.08), as did the indirect effect of spouse constructive accommodation on adjusting activities in response to fatigue and symptoms, via perceived general spouse support (t=1.824, p=.07), In Model B-2, the indirect effect of spouse retaliation on medication adherence via perceived illness-specific spouse support approached statistical significance (t=-1.751, p=.08). Despite significant paths in Model A-2 from spouse constructive accommodation to perceived general support and from perceived general support to adherence to recommended health services, the overall indirect path estimate using Sobel's approach was not significant (t=1.600, p=.11). The same was true in Model B-2 for the effect of spouse retaliation on infection avoidance adherence via illness-specific support (t=-1.412, p=.16) and the effect of spouse retaliation on adjusting activities in response to fatigue or symptoms via illness-specific support (t=-1.508, p=.13).

Discussion

To our knowledge, this study is one of few to investigate specific interaction processes that underlie perceptions of spousal support in the context of chronic illness. It is also the first to investigate how spousal accommodative behavior, an indicator of the construct of transformation of motivation, may influence health behavior in the context of chronic illness. Finally, this study is one of very few to investigate chronic illness self-management among ANCA-SVV patients. Specifically, we investigated whether spouse retaliation and constructive accommodation were associated with perceived spousal support, both general and illness-specific, and if, in turn, perceived spousal support was associated with greater performance of recommended self-management behaviors among patients with ANCA-SVV.

Overall, our results provide partial support for Hypotheses 1 and 2, which assert that spousal accommodative behavior may serve as a relationship process by which perceptions

of spousal support, both general and illness-specific, are determined. As proposed in Hypothesis 2, patients perceived higher levels of general spouse support when they reported that their spouse exhibited higher levels of constructive accommodation, independent of the spouse's level of retaliation and the patient's own self-reported constructive accommodation and retaliation. Contrary to Hypothesis 1, however, higher levels of spouse retaliation did not appear to predict patients' perceived general spouse support, independent of spouse constructive accommodation and the patient's own accommodative behavior. These results suggest that constructive accommodation may be more important than retaliation in determining perceptions of general spousal support. That is, patients' overall perceptions of spousal support may be influenced more by the presence of positive, constructive responses to illness-related conflict than the absence of negative, destructive responses. Interestingly, this pattern of findings was reversed with regard to perceived illness-specific support. That is, spouse retaliation was an independent predictor of patients' perceptions of illness-specific spouse support, as hypothesized, but spouse constructive accommodation was not. Thus, spousal retaliation may be more important than constructive accommodation in determining illness-specific support. It should be noted that this study assessed spouse's accommodative behavior in the context of chronic illness; that is, we examined how spouses respond to patients' destructive behavior *related to their illness* (e.g., when the patient is not feeling well and says something mean to the spouse or withdraws from the spouse). When spouses retaliate in response to these illness-related destructive acts by the patient, patients may feel less supported in terms of their illness specifically, but not less supported by their spouse in general. Future research should evaluate whether spouse retaliation occurring outside of the illness context influences perceptions of general spousal support.

Our results also provided partial support for Hypothesis 3, in that patients' perceptions of both illness-specific and general spousal support were associated with some self-management behaviors, but not others. Specifically, higher levels of both general and illness-specific spouse support predicted higher levels of patients' medication adherence and appropriate adjusting of daily activities in response to illness symptoms and fatigue. In addition, higher levels of general spouse support predicted greater adherence to recommended health services, and higher levels of illness-specific spouse support predicted greater infection avoidance adherence. These results suggest that spouse support may be quite important in facilitating patients' performance of these specific behaviors but less important in determining behavior related to diet, exercise, symptom monitoring, and reporting symptoms and side effects to health professionals. These results are interesting in light of previous research with other illness populations. A recent review of this literature (Gallant, 2003) revealed that, as in this study, higher illness-specific support was associated with improved medication adherence among adults with diabetes and heart disease. This relationship was not found for patients with epilepsy or asthma, and no studies included in the review looked at the relationship of general support to medication adherence specifically. The same review also found that illness-specific support consistently predicted better dietary and exercise adherence among diabetes and heart disease patients, a relationship not found in the current study. Our results, coupled with the results of this recent review, suggest that the relationship among support and performance of self-management behaviors may differ across illnesses, type of support, and specific behaviors.

Much less research has been conducted on the effect of support on several of the other behaviors studied in this current analysis, including adherence to recommended health

services, infection avoidance adherence, and appropriate adjusting of one's activities in response to fatigue and illness symptoms. Our results suggest that general spousal support may be more important than illness-specific support in helping patients to obtain recommended health services, and as important as illness-specific support in helping them to limit daily activities when they feel tired or ill. In order to carry out these behaviors, patients may rely on instrumental help with non-illness-specific daily responsibilities (e.g., child care, household chores) to free them up to attend to the demands of their illness; that is, the type of help they need may be less tied to illness-specific tasks and related more to general life responsibilities. Recent research with the current study sample revealed that many patients feel that work and family responsibilities interfere with their ability to appropriately adjust their activities in response to fatigue and symptoms (Thorpe, *et al*., in preparation); therefore, having a generally supportive spouse who can take on some of these instrumental responsibilities in an emotionally supportive way may in turn facilitate this self-care behavior. In contrast, our results suggest that illness-specific support may be more important than general support with regard to patients' infection avoidance adherence.

The statistical significance of the individual pathways evaluated in these first three hypotheses suggests that perceived support acts as a mediator of the effect of spousal accommodation on patients' performance of several ANCA-SVV self-management behaviors. However, formal assessment of perceived general and illness-specific support as mediators using Sobel's approach did not reveal these indirect paths to be statistically significant. This may be due to the fact that Sobel's product-of-coefficients approach to evaluating indirect effects has been widely documented as having low statistical power, particularly in small

samples (MacKinnon *et al.*, 2002). This is especially plausible given that the indirect effects fell just short of statistical significance at the $p<.05$ level.

The results of the model comparison tests did not support Hypothesis 4. That is, the two models (A-2 and B-2) allowing direct paths from spousal accommodation to the eight self-management variables in addition to indirect paths through perceived spousal support fit the data better than the models without these direct paths. This suggests that while perceived spousal support mediates the relationship between spousal accommodative behavior and several self-management behaviors, as discussed above, it does not do so completely. While with one exception, these overall direct paths were not individually statistically significant, as a group their estimation improved model fit. Furthermore, the only significant direct path (a positive association between spouse retaliation and exercise adherence) was in the opposite direction than expected. It is possible that that these direct effects exist, but were too small to be detected in this study given the small sample size.

Several limitations of this research should be noted. First, the sample size in this study was somewhat small for conducting path analysis. We attempted to offset this limitation as much as possible by carefully choosing model fit indices and estimation methods that have been shown to perform best with small sample sizes (Curran et al., 1996; Hu & Bentler, 1998). However, as noted above, the small sample size may have particularly impacted our ability to detect significant indirect effects, as well as small effects represented by individual direct and indirect pathways in the tested models.

Second, the results of the model fit tests were somewhat contradictory, with the CFI and SRMR suggesting good model fit and the Satorra-Bentler chi-square test suggesting less than acceptable fit. All three of these model fit indices, using the cut-offs used in this study, have

been shown to perform well under conditions of small sample size, multivariate non-normality, and model under-specification (Curran et al., 1996; Hu & Bentler, 1998, 1999), so it is unclear why they provided contradictory information regarding model fit. One possibility is that we had an over-specified model; that is, it was not necessary to control for the large number of socio-demographic and clinical factors in our models. Indeed, many of the correlations of these variables with the main independent variables of interest were non-significant in final models, but post-hoc model modification (e.g., fixing these pathways to zero and evaluating the resulting effect on model fit) is generally not recommended when conducting research with small samples (Hoyle & Panter, 1995). The sensitivity of the model fit indices used in this study to model over-specification has not been extensively investigated and thus we cannot rule it out as a source of the conflicting model fit results. At any rate, the significant Satorra-Bentler chi-square value suggests that we should exercise caution in interpreting the individual parameter estimates produced in the final path models, and future research with larger samples should attempt to replicate the findings of this study.

Third, the cross-sectional nature of the study limits our ability to infer causality. Although causal relationships are implied by the path models tested in this study and the use of the term mediation, it is important to acknowledge that the cross-sectional nature of the study prohibits our ability to infer causal relationships from the parameter estimates produced in the analyses. These analyses should be viewed as exploratory in nature, and significant path coefficients should be viewed as preliminary support for the possibility that independent variables exert a causal effect on dependent variables. Future studies with longitudinal designs are required to begin to make reasonable assertions about causal relationships among the accommodation, support, and self-management constructs.

Fourth, we used a convenience sample of ANCA-SVV patients that may not represent the larger population of ANCA-SVV patients. In particular, because we used a registry of patients with autoimmune kidney-related diseases as a primary recruitment source, our sample may have over-represented patients with kidney involvement. Furthermore, the average disease duration reported by this sample was over six years, and we may have under-represented recently diagnosed individuals. Future research should investigate whether these results can be generalized to all patients with ANCA-SVV, as well as patients with other chronic conditions.

Finally, the self-report measurement used to assess independent, mediating, and dependent variables (e.g., perceived accommodation, perceived support, and perceived self-management behavior) may have inflated the relationships found in this study. Future studies should attempt to replicate these findings using other methods of measuring accommodative behavior (e.g., direct observation, spouse report) and performance of self-management behaviors (e.g., pill counts to assess medication adherence, diary records of diet and exercise behavior).

Despite these limitations, this study provides insight into a specific style of interaction between married couples that may underlie ANCA-SVV patients' perceptions of spousal support, and in turn may influence their performance of several important self-management behaviors. The findings of this study suggest that spousal accommodative behavior may be a promising avenue for interventions to improve illness self-management among ANCA-SVV patients, and potentially patients with other chronic conditions.

Figure 6.1. Typology of accommodative behavior (Rusbult & Verette, 1991).

Active

Exit   Voice

Destructive ————————————————|———————————————— Constructive

Neglect   Loyalty

Passive
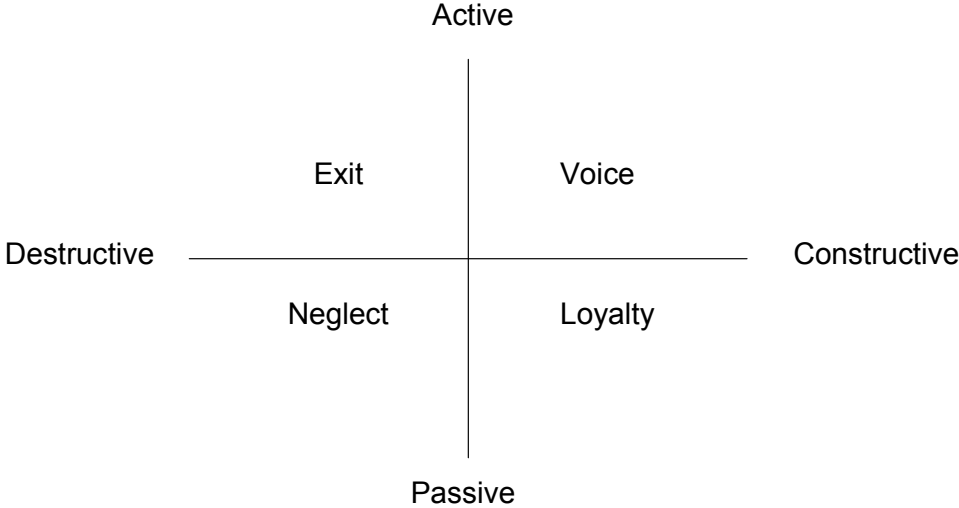
Figure 6.2. Conceptual model of relationships between marital accommodation, perceived spousal support, and patient self-management behavior.
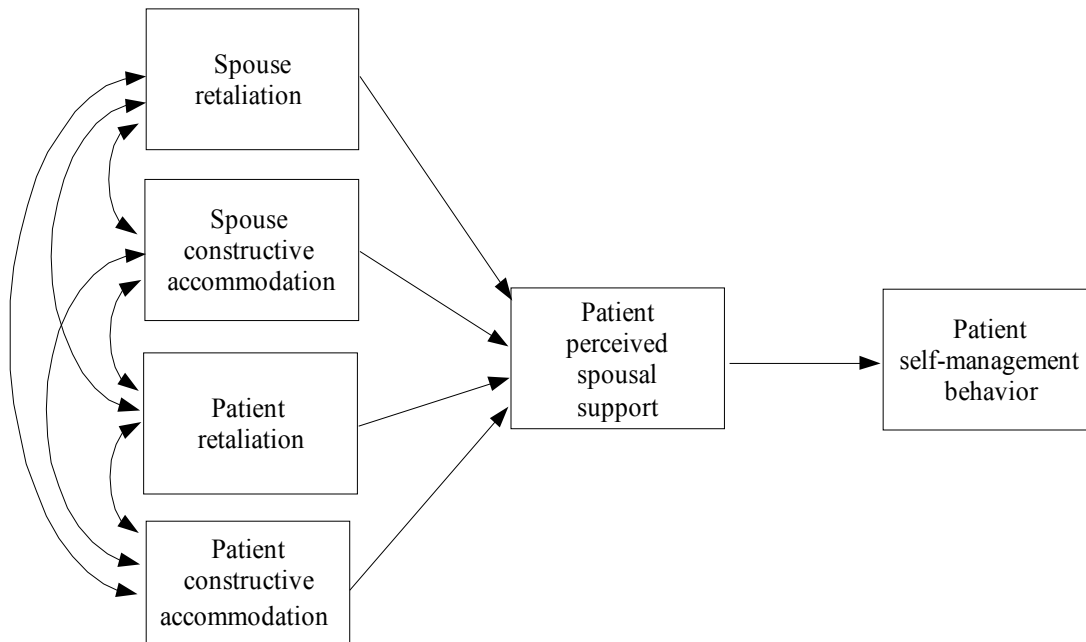
Figure 6.3. Alternative model of relationships between marital accommodation, perceived spousal support, and patient self-management behavior.
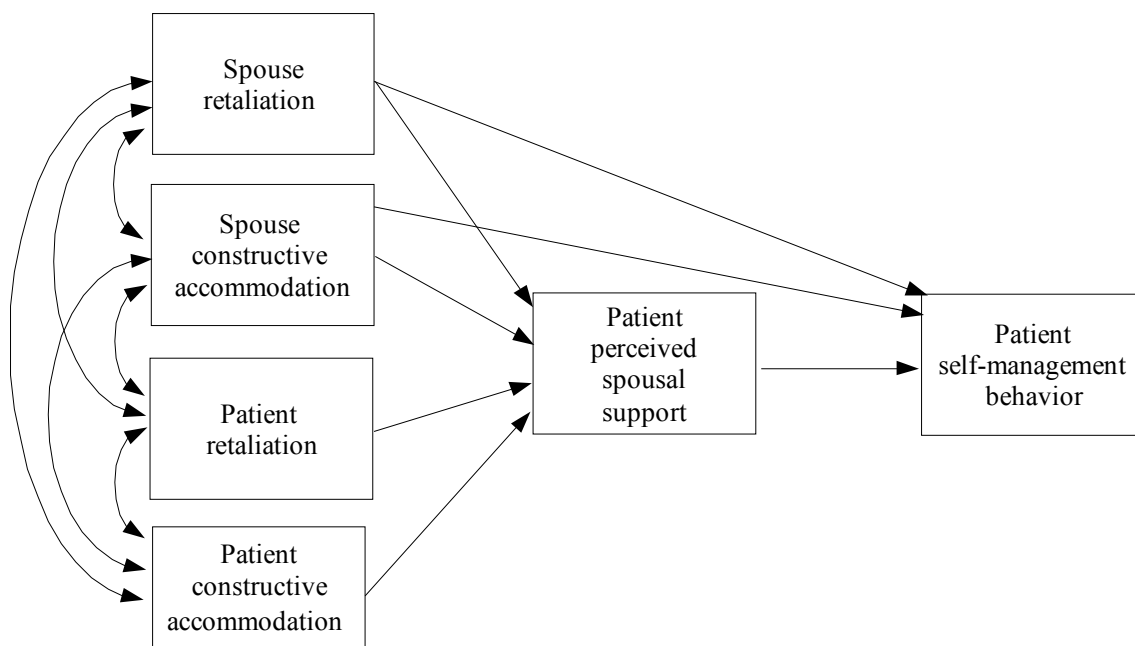
Table 6.1. Descriptive statistics for 159 ANCA-SVV patients and their spouses.

	Cronbach's á	*n*	% or *M (SD)*	Skewness	Kurtosis
Patient Socio-demographics					
Gender					
Male	-	80	50.3%	-	-
Female	-	79	49.7%	-	-
Missing	-	0			
Race/ethnicity					
White	-	151	95.0%	-	-
American Indian/Alaskan Native	-	2	1.3%	-	-
Asian	-	4	2.5%	-	-
Hispanic/Latino	-	1	0.6%	-	-
Missing	-	1	0.6%		
Age, in years	-	159	56.5 (12.8)	-0.13	-0.73
Education level, in years	-	159	14.7 (2.3)	-0.62	-0.75
Duration of marriage, in years	-	159	28.8 (14.9)	0.03	-0.91

Spouse Socio-demographics

Race/ethnicity

White	-	147	92.5%	-	-
American Indian/Alaskan Native	-	1	0.6%	-	-
Asian	-	5	3.1%	-	-
Black or African American	-	1	0.6%	-	-
Native Hawaiian or Other Pacific Islander	-	1	0.6%	-	-
Hispanic/Latino	-	4	2.5%	-	-
Missing	-	0			
Age, in years	-	158	57.7 (12.6)	-0.15	-0.41
Education level, in years	-	159	14.3 (2.4)	-0.74	0.35

Clinical and health characteristics

Diagnosed condition

Wegener's granulomatosis	-	116	73.0%	-	-

Microscopic polyangiitis	-	12	7.6%	-	-
Churg Strauss syndrome	-	9	5.7%	-	-
ANCA-glomerulonephritis	-	22	13.8%	-	-
Missing	-	0			
Ever on dialysis				-	-
Yes	-	20	12.6%		
No	-	136	85.6%		
Missing	-	3	1.9%		
Currently on dialysis (of those ever on dialysis)	-			-	-
Yes	-	4	20.0%		
No	-	16	80.0%		
Missing	-	0	0%		
History of kidney transplant	-		5.8%	-	-
Yes	-	9	5.7%		
No	-	147	92.5%		
Missing	-	3	1.9%		

Time since diagnosis, in months	-	158	77.1 (72.5)	2.22	7.81
Physician rating of vasculitis activity, range 1-10	-	125	2.0 (1.4)	1.88	5.23
Physician rating of vasculitis-related damage, range 1-10	-	108	4.2 (2.3)	0.62	-0.29
Number of recommended adherence behaviors, range 0-8	-	159	5.9 (1.4)	-0.62	-0.28
General physical health, range 0-100	0.81	159	46.8 (23.4)	0.24	-0.89
Psychosocial characteristics					
Perceived spousal support (general), range 24-144	0.89	159	129.8 (14.3)	-1.69	3.30
Perceived spousal support (illness-specific), range 1-6	0.94	157	5.6 (0.7)	-3.29	14.38
Social desirability bias, range 1-20	0.77	159	12.5 (3.7)	-0.05	-0.65
Accommodative behavior					
Spouse constructive accommodation, range 0-8	0.88	155	5.89 (1.46)	-1.11	1.76
Spouse retaliation, range 0-8	0.84	154	1.81 (1.34)	1.00	0.95
Patient constructive accommodation, range 0-8	0.91	152	5.71 (1.38)	-1.06	2.71
Patient retaliation, range 0-8	0.83	152	1.98 (1.27)	0.41	-0.19

Self-management behaviors

Medication adherence, range 1-5	0.77	148	4.5 (0.5)	-0.92	0.25
Adherence to recommended health services, range 1-5	0.71	156	4.6 (0.5)	-2.3	7.15
Infection avoidance adherence, range 1-5	0.86	109	3.8 (0.8)	-0.72	1.30
Dietary adherence, range 1-5	0.75	120	3.6 (0.7)	-0.47	.20
Exercise adherence, range 1-5	0.95	99	3.1 (1.0)	-0.15	-0.73
Symptom monitoring adherence, range 1-5	0.92	104	3.8 (1.0)	-0.27	-0.92
Adjusting activities in response to fatigue or symptoms, range 1-5	0.62	147	3.4 (0.6)	-0.20	0.22
Prompt reporting of illness symptoms and side effects, range 1-5	0.90	132	3.2 (1.1)	-0.03	-0.81

Table 6.2. Bivariable (pairwise) correlations for all variables included in path analysis.

	Gender	Race	Age	Education	Marriage duration	Disease duration	Activity	Damage
Gender	1.00	-	-	-	-	-	-	-
Race	-0.03	1.00	-	-	-	-	-	-
Age	0.30***	0.12	1.00	-	-	-	-	-
Education	-0.16*	-0.02	-0.17*	1.00	-	-	-	-
Marriage duration	0.21**	0.05	0.80***	-0.19*	1.00	-	-	-
Disease duration	0.07	0.03	0.07	-0.02	0.08	1.00	-	-
Vasculitis activity	0.05	-0.02	-0.20*	-0.07	-0.18*	-0.17	1.00	-
Vasculitis damage	0.36***	0.07	0.15	-0.11	0.10	0.13	0.22*	1.00
Number of rec. behaviors	-0.04	0.03	0.07	0.02	0.07	-0.07	0.11	0.21*
General perceived spouse support	0.16*	-0.07	0.12	-0.09	0.06	0.07	0.01	0.08
Illness-specific spouse support	0.24**	-0.04	0.18*	-0.16*	0.08	-0.02	-0.01	0.07

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Social desirability	0.10	-0.13	0.26*	-0.23*	0.26*	-0.02	-0.03	0.01
Spouse constructive behavior	0.15	0.05	0.24*	-0.12*	0.17	-0.07	-0.12	0.15
Spouse retaliation	-0.11	-0.07	-0.09	0.12	-0.04	0.07	0.05	-0.01
Own constructive behavior	0.07	0.06	0.09	-0.04	0.04	-0.04	-0.04	0.15
Own retaliation	-0.17*	-0.08	-0.05	0.18*	0.07	0.07	0.03	-0.11
Medication adherence	0.28***	0.06	0.28*	-0.18	0.23*	-0.11	-0.05	0.04
Health services adherence	-0.10	0.08	-0.02	-0.06	0.05	-0.17*	0.12	-0.09
Infection avoidance adherence	-0.01	-0.11	0.05	0.06	0.13	-0.12	-0.12	-0.03
Diet adherence	0.08	-0.14	0.06	0.13	0.09	0.05	0.05	-0.01
Exercise adherence	0.14	0.01	0.13	0.03	0.15	0.11	0.11	0.03
Symptom monitoring adherence	0.08	-0.02	0.15	-0.15	0.14	-0.05	-0.04	-0.03
Adjusting	0.00	0.07	0.32*	-0.02	0.20*	0.01	-0.21*	0.02

	activity level							
Reporting symptoms	0.05	-0.03	0.02	-0.09	0.06	-0.08	0.11	-0.12

Table 6.2 continued.

	Number of rec. behaviors	General perceived spouse support	Illness-specific spouse support	Social desirability	Spouse constructive behavior	Spouse retaliation	Own constructive behavior	Own retaliation
Number of rec. behaviors	1.00	-		-	-	-	-	-
General perceived spouse support	0.10	1.00		-	-	-	-	-
Illness-specific spouse support	0.01	0.74***	1.00					
Social desirability	-0.01	0.11	0.06	1.00	-	-	-	-
Spouse constructive behavior	0.15	0.46***	0.48***	0.11	1.00	-	-	-
Spouse retaliation	-0.09	-0.39***	-0.52***	-0.23**	-0.61***	1.00	-	-
Own constructive behavior	-0.01	0.27***	0.24**	0.21*	0.60***	-0.27***	1.00	-

Own retaliation	-0.12	-0.54***	-0.46***	-0.19*	-0.44***	0.75***	-0.43***	1.00
Medication adherence	-0.06	0.22**	0.32***	0.23**	0.15	-0.08	0.10	-0.10
Health services adherence	0.07	0.15	0.09	0.11	0.05	-0.13	0.02	-0.09
Infection avoidance adherence	0.06	0.08	0.21*	0.11	-0.01	-0.06	0.18	-0.21*
Diet adherence	0.10	0.11	0.07	0.14	0.06	0.01	0.09	-0.07
Exercise adherence	-0.03	0.06	-0.06	0.21*	0.06	0.06	0.08	0.00
Symptom monitoring adherence	0.14	0.15	-0.05	0.33***	0.19	-0.12	0.13	-0.23*
Adjusting activity level	0.15	0.17	0.16	0.06	0.08	0.01	0.11	-0.12
Reporting symptoms	0.02	0.10	0.17	0.19*	0.06	-0.17	0.13	-0.18*

Table 6.2 continued.

	Medication adherence	Health Services adherence	Infection avoidance adherence	Dietary adherence	Exercise adherence	Symptom monitoring adherence	Adjusting activity level	Reporting symptoms
Medication adherence	1.00	-	-	-	-	-	-	-

Health services adherence	0.22**	1.00	-	-	-	-	-	-
Infection avoidance adherence	0.18	-0.03	1.00	-	-	-	-	-
Diet adherence	0.15	0.05	0.21*	1.00	-	-	-	-
Exercise adherence	0.27**	0.10	-0.02	0.46***	1.00	-	-	-
Symptom monitoring adherence	0.19	0.11	0.08	0.19	0.41***	1.00	-	-
Adjusting activity level	0.12	0.02	0.27**	0.35***	0.03	0.20*	1.00	-
Reporting symptoms	0.32***	0.26**	0.24*	0.11	0.08	0.29*	0.13	1.00

* $p<.05$
** $p<.01$
*** $p<.001$

Table 6.3. Model fit indices for overall fit of all four models and difference in fit for nested models.

	Alternative 1 (support completely mediates relationships betweens spousal accommodation and self-management)			Alternative 2 (allows for direct paths from spousal accommodation to self-management)			Model Comparison
	χ^2			χ^2			χ^2 difference
	(32, 159)	CFI	SRMR	(16, 159)	CFI	SRMR	(16, 159)
Model A	71.386***	0.90	0.03	31.71*	0.96	0.02	39.676***
Model B	65.352***	0.91	0.02	35.692**	0.95	0.02	29.66*

* $p<.05$
** $p<.01$
*** $p<.001$

Table 6.4. Unstandardized beta coefficients and standard errors for primary pathways of interest in Model A-2.

	Spouse support	Medication	Health services	Infection	Diet	Exercise	Symptom monitoring	Adjusting activities	Reporting symptoms & side effects
	b (se)	b (se)	b (se)	b (se)	b (se)	b (se)	b (se)	b (se)	b (se)
Spouse support	--	0.007* (0.003)	0.006* (0.003)	0.002 (0.007)	0.008 (0.006)	0.013 (0.009)	0.007 (0.008)	0.010** (0.004)	-0.006 (0.008)
Spouse retaliation	-0.55 (1.58)	0.05 (0.04)	-0.030 (0.038)	-0.080 (0.082)	0.083 (0.065)	0.202* (0.102)	0.141 (0.090)	0.073[†] (0.044)	-0.186[†] (0.096)
Spouse constructive accommodation	3.36** (1.26)	0.02 (0.04)	-0.015 (0.026)	-0.039 (0.069)	0.057 (0.056)	0.099 (0.115)	0.068 (0.078)	-0.012 (0.032)	-0.009 (0.80)
Own retaliation	-4.82** (1.26)	--	--	--	--	--	--	--	--
Own constructive accommodation	-1.25 (0.93)	--	--	--	--	--	--	--	--
Gender	0.22 (1.84)	0.21* (0.09)	-0.104 (0.085)	-0.016 (0.184)	0.097 (0.157)	0.220 (0.240)	0.196 (0.221)	-0.103 (0.108)	0.207 (0.194)
White	-8.09 (5.33)	-0.02 (0.02)	0.307 (0.212)	-0.428 (0.435)	-0.314 (0.293)	0.827* (0.407)	-0.112 (0.392)	0.131 (0.171)	0.021 (0.377)
Age	0.03 (0.11)	0.005 (0.005)	-0.008 (0.007)	-0.008 (0.010)	-0.005 (0.008)	0.003 (0.017)	0.000 (0.011)	0.018** (0.006)	-0.006 (0.012)

Education	0.31 (0.41)	-0.020 (0.018)	-0.009 (0.017)	0.018 (0.033)	0.057* (0.027)	0.054 (0.045)	-0.040 (0.039)	0.000 (0.020)	0.004 (0.041)
Marriage duration	0.03 (0.09)	0.001 (0.004)	0.007 (0.006)	0.011 (0.009)	0.007 (0.006)	0.000 (0.014)	-0.004 (0.008)	-0.005 (0.004)	0.009 (0.010)
Duration of disease	0.03** (0.01)	-0.001† (0.001)	-0.001† (0.001)	-0.001 (0.001)	0.001 (0.001)	0.002 (0.001)	0.001 (0.001)	0.000 (0.001)	0.000 (0.001)
Disease activity	1.07 (0.71)	0.013 (0.033	0.034 (0.030)	-0.051 (0.065)	0.064 (0.050)	0.133 (0.089)	-0.067 (0.080)	-0.075† (0.045)	0.147* (0.074)
Disease damage	0.001 (0.49)	-0.018 (0.023)	-0.016 (0.027)	-0.006 (0.043)	-0.026 (0.033)	-0.071 (0.054)	-0.052 (0.047)	0.008 (0.029)	-0.113* (0.049)
Total number of behavioral recommendations	0.06 (0.84)	-0.028 (0.031)	0.016 (0.026)	0.078 (0.087)	0.048 (0.064)	-0.011 (0.109)	0.180 (0.084)*	0.046 (0.034)	0.072 (0.065)
Social desirability bias	-0.04 (0.28)	0.026* (0.013)	0.015 (0.010)	0.004 (0.026)	0.026 (0.018)	0.078** (0.028)	0.068 (0.078) **	0.004 (0.013)	0.049† (0.026)
Intercept	124.294 (11.762)	2.920** (0.739)	3.958** (0.581)	3.855** (1.299)	0.877 (1.000)	-2.339 (1.801)	1.009 (1.463)	0.976 (0.742)	3.367** (1.392)

† $p<.10$
* $p<.05$
** $p<.01$

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Table 6.5. Unstandardized beta coefficients and standard errors for primary pathways of interest in Model B-2.

	Spouse support	Medication	Health services	Infection	Diet	Exercise	Symptom monitoring	Adjusting activities	Reporting symptoms & side effects
	b (se)	b (se)	b (se)	b (se)	b (se)	b (se)	b (se)	b (se)	b (se)
Spouse support	--	0.232** (0.064)	0.059 (0.071)	0.277* (0.139)	0.180† (0.108)	0.097 (0.187)	-0.190 (0.161)	0.195* (0.085)	0.103 (0.171)
Spouse retaliation	-0.180* (0.090)	0.067† (0.040)	-0.041 (0.044)	-0.027 (0.083)	0.089 (0.067)	0.170 (0.109)	0.072 (0.095)	0.075 (0.046)	-0.140 (0.099)
Spouse constructive accommodation	0.109 (0.068)	0.014 (0.036)	-0.007 (0.028)	-0.063 (0.065)	0.059 (0.067)	0.121 (0.111)	0.102 (0.078)	-0.008 (0.033)	-0.033 (0.079)
Own retaliation	-0.058 (0.063)	--	--	--	--	--	--	--	--
Own constructive accommodation	-0.010 (0.047)	--	--	--	--	--	--	--	--
Gender	0.145 (0.091)	0.177* (0.089)	-0.108 (0.083)	-0.057 (0.184)	0.078 (0.154)	0.218 (0.244)	0.232 (0.218)	-0.122 (0.106)	0.186 (0.199)
White	-0.328 (0.271)	0.279 (0.203)	0.279 (0.216)	-0.354 (0.418)	-0.321 (0.294)	0.753† (0.417)	-0.226 (0.394)	0.113 (0.169)	0.101 (0.341)
Age	0.011* (0.005)	0.003 (0.005)	-0.008 (0.007)	-0.012 (0.011)	-0.006 (0.008)	0.003 (0.018)	0.003 (0.011)	0.016** (0.006)	-0.008 (0.012)

Education	-0.016 (0.019)	-0.015 (0.018)	-0.008 (0.018)	0.023 (0.033)	0.060* (0.027)	0.057 (0.046)	-0.044 (0.039)	0.004 (0.020)	0.006 (0.041)
Marriage duration	-0.005 (0.004)	0.002 (0.004)	0.007 (0.006)	0.012 (0.008)	0.007 (0.006)	0.000 (0.014)	-0.005 (0.009)	-0.004 (0.004)	0.010 (0.010)
Duration of disease	0.000 (0.001)	-0.001 (0.001)	-0.001 (0.001)	-0.001 (0.001)	0.001 (0.001)	0.002 (0.001)	0.002 (0.001)	0.000 (0.001)	0.000 (0.001)
Disease activity	0.053 (0.041)	0.009 (0.031)	0.036 (0.031)	-0.064 (0.062)	0.062 (0.050)	0.141 (0.090)	-0.051 (0.079)	$-0.076^{\dagger}$ (0.043)	$0.136^{\dagger}$ (0.074)
Disease damage	-0.007 (0.028)	-0.016 (0.022)	-0.015 (0.028)	-0.004 (0.042)	-0.024 (0.033)	-0.069 (0.055)	-0.052 (0.047)	0.010 (0.029)	-0.113 (0.049)*
Total number of behavioral recommendations	-0.028 (0.043)	0.002 (0.004)	0.020 (0.026)	0.086 (0.085)	0.056 (0.064)	-0.002 (0.110)	0.178* (0.083)	0.055 (0.035)	0.072 (0.063)
Social desirability bias	-0.020 (0.015)	0.031** (0.013)	0.015 (0.010)	0.009 (0.025)	0.029 (0.019)	0.079** (0.027)	0.079** (0.025)	0.007 (0.014)	0.052 (0.025)*
Intercept	5.792** (0.712)	2.489** (0.763)	4.331** (0.705)	2.495 (1.431)	0.829 (1.050)	-1.321 (1.692)	$2.891^{\dagger}$ (1.545)	1.077 (0.770)	2.077 (1.438)

† $p<.10$
* $p<.05$
** $p<.01$

Table 6.6. Test statistics for Sobel tests for significant indirect effects of spousal accommodative behavior on self-management, through perceived social support.

	Medication	HS	Infection	Diet	Exercise	Sym Mon	Adjusting	Reporting
Model A-2								
Spouse retaliation	-0.344	-0.343	-0.221	-0.337	-0.338	-0.323	-0.345	0.316
Spouse constructive	1.756†	1.600	0.284	1.193	1.270	0.831	1.824†	-0.722
Model B-2								
Spouse retaliation	-1.751†	-0.767	-1.412	-1.280	-0.502	1.016	-1.508	-0.577
Spouse constructive	1.466	0.738	1.249	1.155	0.494	-0.950	1.314	0.564

$^\dagger p<.10$

CHAPTER SEVEN: SUMMARY AND DISCUSSION

In this chapter, I summarize the primary findings of the dissertation and discuss implications of these findings for ANCA-SVV self-management and chronic illness management more generally. The major limitations and strengths of the study are then discussed, along with directions for future research.

Summary of findings

The first manuscript, presented in Chapter 4, reports on the development and initial evaluation of the Vasculitis Self-management Scale (VSMS). As expected, self-management behavior in ANCA-SVV patients was found to be multi-dimensional. Specifically, eight domains of behavior were identified as relevant for the majority of individuals in the study sample, including six adherence domains (medication, health services, infection avoidance, diet, exercise, and symptoms monitoring adherence) and two general self-care domains (prompt reporting of new symptoms and side effects to a heath professional and appropriate adjusting of activities in response to fatigue or symptoms). The final version of the VSMS assessed each of these eight domains in separate subscales. As expected, individuals' performance of each self-management behavior was relatively independent of their performance of the other behaviors, as evidenced by null to relatively modest Pearson correlations between scores on the eight subscales. While participants' performance of some self-management behaviors was significantly correlated to their performance of several other behaviors (e.g., dietary and exercise adherence were significantly correlated), the largest of these observed correlations did not exceed $r=0.37$. Other

analyses in this chapter demonstrated acceptable internal consistency and test-retest reliability for the eight VSMS subscales. Cronbach's $\acute{\alpha}$ for the subscales ranged from minimally acceptable for the adjusting activity level subscale ($\acute{\alpha} = 0.67$) to excellent for the exercise adherence subscale ($\acute{\alpha} = 0.94$), while test-retest correlations ranged from $r = 0.56$ for adherence to recommended health services to $r = 0.79$ for exercise adherence.

Results for the final set of analyses reported in Chapter 4 examining the construct validity of the VSMS were more mixed in nature. Convergent validity was supported by the direction and magnitude of the observed correlations of many of the VSMS subscales (in particular, the subscales for medication adherence, diet adherence, symptom monitoring adherence, health services adherence, and prompt reporting of symptoms and side effects) with the psychosocial and behavioral variables (e.g., general adherence, social support, self-efficacy, and depressive symptoms), although not all correlations were significant as expected. The non-significant correlations of age and gender with most of the VSMS subscales also provided some evidence of discriminant validity, although unexpected significant correlations were found for medication adherence and gender, as well as for age and three of the subscales (medication adherence, diet adherence, and adjusting activities in response to fatigue and symptoms). We did not observe the expected relationships between the VSMS subscales and the clinical (e.g., ratings of disease activity and damage) and regimen characteristics (e.g., ratings of treatment complexity, number of medications, and number of behavioral recommendations). We also observed unexpected results for the relationship between education and scores on the self-management subscales, which were either null or in the opposite direction of our hypotheses, as well as for scores on the social desirability scale with scores on five of the eight self-management subscales, which were significant, positive, and modest in magnitude. Whether these correlations did not conform to

expectations due to poor validity of the VSMS, poor validity or reliability of the chosen validation measures, or a lack of a true relationship between these characteristics and self-management behavior in ANCA-SVV patients is a question for future research (discussed in more detail below).

The second manuscript, presented in Chapter 5, characterized patient attitudes and beliefs about performing ANCA-SVV self-management behaviors. Our results suggest that among the self-management challenges they face, adults with this condition experience the least difficulty in using recommended health services and taking medication. Patients reported the highest levels of adherence, the highest levels of perceived importance, and greatest number of perceived facilitators for these behaviors. They also rated these behaviors lowest with regard to perceived difficulty. Analysis of these same indicators revealed that patients appeared to have the most difficulty with exercise adherence, adjusting activities in response to fatigue or symptoms, and prompt reporting of symptoms and side effects. Patients also perceived dietary recommendations as difficult and less important to follow, although mean levels of actual dietary adherence were moderate. We also found that several patient perceptions were related to actual performance of these behaviors. Both perceived difficulty of performing the behavior and the total number of barriers that participants reported for each behavior tended to be negatively associated with scores on the corresponding VSMS subscale, while greater perceived importance of the behavior tended to be positively associated with VSMS scores. Finally, the research reported in Chapter 5 also identified specific perceived barriers and facilitators to ANCA-SVV self-management that are salient to our sample of ANCA-SVV patients and may affect their performance of self-management behaviors. Across behaviors, perceived benefits (e.g., a belief that performing the behavior is very important for maintaining good health) were frequently

mentioned as facilitating many of the behaviors assessed by the VSMS, although reporting this facilitator was not significantly associated with higher levels of self-management. With the exception of perceived benefits, the barriers and facilitators most frequently mentioned by patients varied substantially across behaviors, as did significant relationships between reporting of these barriers and facilitators and self-management behavior. Patients' reporting of several specific barriers, but not specific facilitators, was significantly and negatively associated with their score on the corresponding VSMS subscale. Many of these perceived barriers reflected factors found to be associated with self-management in other illness populations; however, the results in Chapter 5 shed light on barriers to self-management behaviors not often studied in other illness contexts, as well.

The research presented in the third manuscript in Chapter 6 provided preliminary support for the hypothesis that spousal accommodative behavior may determine perceptions of spouse support, both general and illness-specific, among adults diagnosed with ANCA-SVV; however, relationships among spousal accommodative behavior and perceived spouse support differed by the type of perceived support assessed. Higher levels of spouse retaliation were associated with lower levels of illness-specific perceived spouse support, but this relationship did not hold true for general perceived spouse support. In addition, higher levels of spousal constructive accommodation were associated with higher levels of general perceived spouse support, but this relationship did not hold true for illness-specific support. We also found partial support for our hypotheses that both illness-specific and general spouse support are associated with self-management behavior. Specifically, higher levels of both general and illness-specific spouse support were associated with higher levels of patients' medication adherence and appropriate adjusting of activities in response to fatigue and symptoms. In addition, higher levels of general

spouse support were associated with greater adherence to recommended health services, and

higher levels of illness-specific spouse support predicted greater infection avoidance adherence.

Taken together, these results suggest that spousal accommodative behavior may determine

patients' perceptions of spouse support, which in turn may determine their performance of at

least some self-management behaviors; however, formal tests of these mediational pathways fell

short of statistical significance.

Implications

The findings summarized above have a number of implications with regard to ANCA-SVV

specifically, as well as chronic illness self-management more generally.

Multi-dimensionality of Illness Self-management

This study revealed that most adults living with ANCA-SVV, on average, are required to

perform a number of behaviors to self-manage their condition, although the specific behaviors

vary to some degree from patient to patient. We also found that correlations among patients'

reports of their performance of each behavior were, at most, small-to-moderate in magnitude,

which is consistent with prior research in other illness populations (Toobert & Glasgow, 1994;

Johnson, 1992; WHO, 2003). In addition, levels of self-management behaviors, as well as

patients' attitudes and beliefs about specific behaviors, varied substantially across the behaviors

assessed in this study. Considered together, these results strongly point to the importance of

considering specific illness self-management behaviors, and their influences, individually. That

is, measures of self-management behavior should assess these behaviors separately instead of

attempting to gather information on one's global adherence to all behavioral recommendations,

and interventions should develop behavior-specific strategies versus trying a "one-size-fits-all"

approach to self-management behavior change. This appears to be true for both ANCA-SVV

and other chronic illnesses.

Usefulness of the VSMS in Assessing ANCA-SVV Self-management

The results of the factor analysis, reliability assessment, and construct validity evaluation in

Chapter 4 suggest that the VSMS is a promising tool for assessing the specific behaviors most

relevant for the self-management of ANCA-SVV. While some of the results of the construct

validity evaluation were discouraging, further evidence of the construct validity of the VSMS

was provided, indirectly, in Chapters 5 and 6. That is, the observed relationships between patient

attitudes and beliefs, including their reporting of specific barriers and VSMS scores in Chapter 5,

and between perceived spousal support and VSMS scores in Chapter 6, provide further evidence

of construct validity of the VSMS. Thus, future research and intervention work regarding

ANCA-SVV self-management should strongly consider using the VSMS, while employing

additional methods for assessing its validity (discussed further below).

Explanations for the Mixed Evidence for Construct Validity of the VSMS

With this in mind, the mixed results for construct validity in Chapter #4 should not be ignored,

as they suggest three important possibilities: 1) the VSMS subscales are not valid measures of

ANCA-SVV self-management behavior; 2) the measures used to assess the construct validation

variables are invalid or unreliable themselves; and/or 3) true relationships between the validation

constructs and specific ANCA-SVV self-management behaviors differ from those implied by

prior research in other illness contexts. Given the other support for construct validity provided

by this research, the latter two explanations for the mixed pattern of results are highly possible.

Indeed, we tended to observe the expected, significant relationships for variables assessed using

established measures (e.g., General Adherence Scale, Social Provisions Scale, CES-D), and

tended to observe unexpected, null relationships for variables assessed using measures with unknown psychometric properties, although this pattern did not always hold true. The last possibility-- that relationships of several of the socio-demographic, clinical, regimen, and psychosocial variables to self-management behavior may vary across specific behaviors and/or illnesses-- is intriguing and has not been well-tested in the chronic illness literature (Gallant, 2003). In fact, the literature on treatment adherence and self-management often acknowledges that behavioral domains are relatively independent of one another, but then discusses models of adherence/self-management and their influences globally, as though they were unidimensional constructs (e.g., WHO, 2003). A more fruitful and useful approach might be to test the applicability of these various models across different specific self-management behaviors (e.g., medication adherence versus infection avoidance adherence) and illnesses (e.g., ANCA-SVV versus diabetes or heart disease), and adapt them as indicated.

One additional finding from the construct validity evaluation is worth noting again here. Contrary to expectations, social desirability, which was measured using a well-established measure, was found to be modestly correlated with several VSMS subscales. Prior research in other illness populations has also routinely found patients to be positively biased in their self-report of self-management behavior (Rand, 2000; Hays, 1993). In light of this evidence, it is probably prudent to take into account (e.g., via statistical controls) social desirability when studying self-management behavior with the VSMS, and when using other self-report measures of illness self-management behavior as well.

Potential Use of VSMS Subscales with Other Illness Populations

While this research focused on the self-management behaviors relevant for adults living with ANCA-SVV, all of these behaviors are relevant for a variety of chronic illnesses, including

several that are very understudied in the self-management and treatment adherence literature.
These behaviors, which are particularly relevant for other relapsing, remitting illnesses and those
treated with immunosuppressive medications (e.g., lupus, rheumatoid arthritis, cancer), include
infection avoidance adherence, symptom monitoring adherence, adjusting activities in response
to fatigue and symptoms, and prompt reporting of symptoms and side effects. Because the
VSMS was developed with flexibility of use with a variety of patients at different stages of the
disease as a primary goal, it may also be appropriate to use its subscales with patients with other
relapsing, remitting, autoimmune diseases. Researchers attempting to study self-management in
these illnesses should consider adapting and using the VSMS subscales to assess these behaviors.

Directions for Intervention with ANCA-SVV Patients

Targeting Specific Barriers and Facilitators for Each Self-management Behavior

In Chapter 5, a number of specific barriers and facilitators that ANCA-SVV patients perceive
in relation to performing self-management behaviors were identified. Examination of these
factors, particularly those barriers that were significantly associated with actual self-management
behavior, suggests promising avenues for intervention development with these patients.

Among the self-management behaviors that have been extensively studied with other patient
populations, including medication adherence, adherence to recommended health services, dietary
adherence, and exercise adherence, many of the most frequently reported barriers and facilitators
in this study have been previously documented as influences on adherence. For example,
routinization of one's daily activities, and incorporating medication-taking into this routine, have
been previously found to facilitate medication adherence among individuals living with HIV
(Ryan & Wagner, 2003; Wagner & Ryan, 2004), a disease in which medication regimens can

also be complex. Similarly, the use of pillboxes for organizing medication regimens has been associated with improved medication adherence among individuals living with HIV (Kalichman *et al.*, 2005). As with prior research in other rheumatic disease populations (Garcia Popa-Lisseanu *et al.*, 2005), a high proportion of patients in this study indicated that the difficulty or ease with which they could schedule appointments at preferred times played a key role in their ability to adhere to recommendations for using health services, although reporting of this as a barrier or facilitator was not related to self-reported adherence. Although not significantly related to adherence to recommended health services in this study, geographic distance from health care providers was also frequently mentioned as a barrier to attending appointments with health professionals, just as greater geographic distance from health care facilities has been documented as a negative influence on utilization of outpatient care in other patient populations (McCarthy & Blow, 2004). Likewise, the specific barriers mentioned by patients and associated with worse dietary adherence, namely frequent exposure to prohibited foods, food preferences, emotional eating, and low motivation to adhere, have been found to be powerful predictors of dietary behavior in other patient populations (Schultz *et al.*, 2001; Yancy & Boan, 2006). Finally, lack of motivation, time, facilities, and someone with whom to exercise have all been associated with reduced physical activity levels in prior research (Dominick & Morey, 2006) and are reflected in this study as well. The similarities of the perceived barriers to these specific self-management behaviors in ANCA-SVV patients compared to other chronically ill individuals suggests that researchers and clinicians may want to look to interventions that have been used successfully with other patients for guidance in developing intervention strategies for ANCA-SVV patients.

As noted above, much less research has been conducted on the remaining four self-management behaviors assessed in this study: adherence to recommended infection avoidance behaviors, symptom monitoring adherence, adjusting activity level in response to fatigue or symptoms, and prompt reporting of illness symptoms and side effects. In this study, the barriers reported for infection avoidance adherence revealed that patients' desire to carry on with their normal activities, whether they are work-related, family-related, or social/leisure-related, were perceived to conflict with their adherence, although only the belief that work responsibilities got in the way was significantly related to adherence. Patients who perceived higher levels of illness-specific spouse support (but not general spouse support) also reported higher levels of infection avoidance adherence. With regard to adjusting activities in response to fatigue or symptoms, respondents once again commonly perceived that their responsibilities, both work and family related, made it difficult to obtain the rest their bodies needed; on the flip side, others perceived reduced or flexible work schedules and retirement as a key facilitator. Many respondents also reported reluctance in reducing their activities or asking for help because of worries about others' reactions to them doing so. Furthermore, patients who perceived higher levels of general spouse support (but not illness-specific support) tended to more appropriately adjust their activities in response to fatigue or symptoms. These results suggest that patients' ability to perform these two self-management behaviors may be heavily influenced by the social resources available to them. Interventions targeted at improving social resources and providing patients with additional instrumental support (from either formal or informal sources) may be particularly warranted.

With regard to symptom monitoring adherence, simple forgetting and low perceived benefits (i.e., a belief that symptom monitoring was not necessary for controlling one's condition) were

commonly reported, and reporting of these perceptions predicted worse adherence.

Intrapersonal-level interventions aimed at increasing perceived benefits and improving patients'

skill in remembering to check symptoms regularly might be particularly effective in improving

this type of adherence.

Finally, prompt reporting of new or increased illness symptoms and side effects appears to be

a particularly difficult issue for many patients, as indicated by the frequency with which a

number of barriers to this action were endorsed by patients. Most notably, patients reported that

their uncertainty as to whether symptoms are related to vasculitis or its treatment or are

meaningful enough to report often served as barriers to prompt reporting. Many patients were

wary of going through the trouble of trying to speak with a health professional or "bothering" the

health professional about something that might not be important, and were inclined to wait and

see if the symptom would resolve on its own. Endorsement of these barriers was in turn related

to greater delay in reporting of symptoms and side effects. These findings suggest that

interventions targeting providers in addition to patients may be necessary for improving patients'

reporting of new symptoms and side effects. For example, providers could be trained to more

explicitly encourage patients to report changes they notice, and their offices could be restructured

to implicitly encourage patients to do so as well (e.g., provide several methods for getting in

touch with providers instead of relying on voice messages only).

Leveraging Spousal Influences on Self-management

Our findings in Chapter 6 suggest that spouse support may play an important role in

influencing some domains of self-management, including medication adherence, health services

adherence, infection avoidance adherence, and appropriate adjusting of activities in response to

fatigue and symptoms. Furthermore, our results suggest that spousal accommodative behavior may be one leverage point for influencing patients' perceptions of spouse support, an endeavor that has proved to be difficult in prior intervention research (e.g., Park, Schultz, et al., 2004; Lichtenstein, Glasgow, et al., 1986). In particular, decreasing spousal retaliation may influence perceptions of illness-specific support, and increasing spousal constructive accommodation may influence perceptions of general support. The development of interventions for patients and their spouses that teach both partners these more adaptive ways of dealing with conflict and dissatisfaction in their relationship may yield improvements in patients' perceptions of support and self-management behavior, in addition to possible improvements in relationship outcomes suggested by prior observational research (Rustbult, Johnson, et al., 1986; Rusbult, Verette, et al., 1991; Wieselquist, Rusbult, et al., 1999).

Overall Study Limitations and Strengths

There are several important limitations that apply to all three studies described in this dissertation. First, the sample consisted of a convenience sample of ANCA-SVV patients that may not represent the larger population of ANCA-SVV patients. In particular, because we used a registry of patients with autoimmune kidney-related diseases as a primary recruitment source, our sample may have over-represented patients with kidney involvement. Furthermore, the average disease duration reported by this sample was over six years, and we may have under-represented recently diagnosed individuals. However, the socio-demographic profile of our patient sample was similar to those reported for other United States samples of adults with systemic vasculitis (Carruthers, et al., 1996; Hoffman, et al., 1998).

Second, the sample used in this research was relatively small, especially when missing data was taken into account. However, in each manuscript, we attempted to minimize reductions in

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power due to the small sample size through the use of various strategies for handling missing data (e.g., using the pairwise correlation matrix in Chapter 4 and multiple imputation in Chapter 6) that use all available data, in contrast to the more conventional listwise deletion. In addition, given the rarity of ANCA-SVV, our sample size was actually quite large compared to samples used in other studies of the psychosocial aspects of this condition.

Third, we relied on patient recall for assessing which self-care behaviors had been explicitly recommended to them by a health professional, as well as their actual performance of recommended behaviors. Thus, the six adherence subscales are actually measuring *perceived* adherence. This could result in the inflation of scores for these six subscales, due to the exclusion of patients not recalling that a behavioral recommendation had been made (and presumably, not performing the behavior as recommended). Missing data due to patients' failure to recall their physicians' recommendations could also have biased parameter estimates for relationships between our independent variables of interest and the VSMS scores in all three papers, although the direction of this bias is difficult to predict. In addition, using patient recall to assess performance of self-management behaviors along with many of our primary independent variables of interest (e.g., perceived barriers, perceived spousal support) may have inflated relationships between these variables.

Fourth, we relied on cross-sectional data for our analyses, yet the research questions examined in Chapters 5 and 6 were causal in nature. While theory and our hypotheses suggest that patient attitudes and beliefs about self-management, spousal accommodative behavior, and perceived spouse support exert a causal effect on self-management behavior, the cross-sectional nature of our study does not help us rule out the possibility that self-management behavior influences these variables, or that an unobserved confounding variable is responsible for the observed

relationships. The cross-sectional design of the research was deemed appropriate and acceptable given the complete lack of prior research on these topics, but the analyses presented in both of these chapters should be considered exploratory in nature and as preliminary support for the possibility that independent variables exert a causal effect on self-management behavior.

Fifth, as noted above, the measurement of several of our independent and control variables was less than ideal. Specifically, patient attitudes and beliefs about self-management, including specific perceived barriers and facilitators, as well as many of the regimen and clinical characteristics, were assessed using single items with unknown psychometric properties. Furthermore, we used open-ended items to assess specific perceived facilitators, which may have placed a high burden on respondents and led to under-reporting of facilitators, and thus, attenuated relationships between specific perceived facilitators and VSMS scores.

This research also has several important strengths worth emphasizing again here. First, we have developed a convenient, flexible tool for assessing self-reported self-management behavior among ANCA-SVV patients. The self-report nature of the VSMS also allows for easy assessment of self-care behaviors not conveniently measured using objective measures. Thus, the development and initial evaluation of the VSMS will greatly facilitate future investigations on this topic. Second, the VSMS may also prove useful for assessing specific self-management behaviors relevant in other illnesses that have been relatively understudied (e.g., infection avoidance adherence, prompt reporting of symptoms and side effects). Third, this is the first investigation, to our knowledge, that examines influences on self-management among adults living with ANCA-SVV. Thus, it serves as an important first step in developing interventions to improve self-management among this understudied patient population. Finally, this study is also, to our knowledge, the first to examine the effect of spousal accommodative behavior on

perceived support and self-management among chronically ill individuals. Thus, it adds significantly to our knowledge about how spouses may influence chronic illness self-management.

Directions for future research

Both the implications and limitations discussed above suggest a number of directions for future research on chronic illness self-management generally and with regard to ANCA-SVV.

First, more research is needed with ANCA-SVV patients to further assess the validity of the VSMS subscales. Ideally, this research would employ different methods than our approach to assessing construct validity. For example, comparing scores on the VSMS subscales to objective measures of adherence (e.g., medication levels in blood, clinic records documenting patients' use of health services) would help evaluate the measure's predictive validity. Examining correlations of scores on the VSMS subscales to objective measures of disease control (blood and protein levels in urine, ANCA titers, or kidney function) or complications (e.g., incidence of infections) would also help evaluate the measure's validity. This research may also want to attempt to identify threshold scores on the VSMS subscales that indicate clinically significant negative outcomes, such as increases in disease activity, new disease damage, or declines in emotional health and quality of life (i.e., at what level of non-adherence as measured by the VSMS subscales are significant adverse health outcomes first seen?). In addition, more research is also needed to evaluate rates of unintentional non-adherence that might be missed by the VSMS because of its reliance on patient recall of behavioral recommendations. That is, it would be valuable to compare physicians' reports of the behavioral recommendations they have made to patients to patients' own recall of these recommendations as they report on the VSMS.

Second, future research should explore the psychometric properties of the VSMS subscales after adapting it for use with other patient populations. As noted above, many or all of the subscales may be appropriate for use with patients diagnosed with other relapsing, remitting, and/or autoimmune conditions, such as lupus, arthritis, or cancer; however, evidence of the measure's factor structure, validity, and reliability when used in this way must be evaluated before concluding this is the case.

Third, as implied above, future research should investigate whether relationships of socio-demographic, clinical, regimen, and psychosocial variables, including perceived support, to self-management behavior vary across specific behaviors and/or illnesses. These variables are widely touted as influencing adherence and self-management globally, when indeed there are indications that they may differentially influence specific self-management behaviors, or that they may be more or less important in some diseases versus others.

Another fruitful line of research would build on our findings presented in Chapter 5 about the specific perceived barriers and facilitators that patients feel affect their performance of specific self-management behaviors. As noted above, our measurement of patient perceptions was less than ideal. Future studies should investigate the factors identified in our study as potentially important to self-management behavior using more established measures of these factors (e.g., does physicians' use of patient-centered communication strategies predict patients' reporting of symptoms and side effects?). Studies employing longitudinal designs would be particularly informative.

The research presented in Chapter 6 also suggests several directions for future investigations. Because of the exploratory nature of these analyses, it would be informative to test the models of spousal accommodative behavior, perceived spouse support, and patient self-management found

to best fit the data in this study using longitudinal data with larger samples of ANCA-SVV patients, as well as with other chronically ill populations. As in this study, these models should evaluate both indirect effects of spousal accommodative behavior on patients' self-management via its influence on perceived spouse support, as well as direct influences of spousal accommodative behavior on patients' self-management. In addition, intervention development work is also needed to explore how spousal accommodative behavior might be changed via couples' interventions, and if doing so influences levels of perceived support. Finally, our findings, which tie specific patterns of spousal interaction to perceptions of spouse support, provide empirical support for taking a relationship processes perspective (Reis & Collins, 2000) when studying social support in the context of chronic illness. Future research should investigate other potential influences on patients' perceptions of social support, both from spouses and other close social network members, to identify other promising avenues for interventions.

Conclusion

This research is the first to examine illness self-management among adults living with ANCA-SVV, a rare, relapsing and remitting autoimmune disease. Our development of the Vasculitis Self-Management Scale (VSMS), and gathering of data regarding its reliability and validity, should facilitate future investigations of self-management among these patients. In addition, our elucidation of the specific barriers and facilitators that ANCA-SVV patients perceive as influencing their illness self-management suggests several promising avenues for future intervention-development efforts to improve self-management in this population. Our findings also suggest that spousal accommodative behavior may influence ANCA-SVV patients' perceptions of spouse support, and thus, their performance of several self-management behaviors;

thus spousal accommodative behavior might be a particularly important target point for future

interventions with this understudied population.

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