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HEALTH RELATED HARDINESS AND PSYCHOSOCIAL ADAPTATION IN
INDIVIDUALS WITH INHERITED BLEEDING DISORDERS AND OTHER
CHRONIC ILLNESSES

A DISSERTATION SUBMITTED TO THE GRADUATE DIVISION OF THE
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This project is dedicated to my parents, Raymond and Genevieve Vasquez.

Te Amo!
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ABSTRACT

An individual who is diagnosed with an inherited bleeding disorder is expected to manage his or her condition on a daily basis. This chronic situation can totally disrupt psychosocial functioning and make it more difficult to adjust to the illness. Other researchers have studied this phenomenon in various other chronic illnesses; however, not in individuals with inherited bleeding disorders (Akkasilpa, et al, 2000, Pollack, 1989a, 1989b). Psychosocial problems are not restricted to individuals with one chronic illness and clinically, it is noted that some individuals adjust to chronic diseases better than others. Individuals living with inherited bleeding disorders may also have other chronic illnesses such as hypertension, asthma, diabetes mellitus (DM), congestive heart failure (CHF), arthritis, and hepatitis A, B, C and/or HIV. The aims of this study are to describe health stressors, health related hardiness, perception of illness impact, self perception of health status and psychosocial adjustment to illness in individuals living with an inherited bleeding disorder; to determine relationships between demographic and illness variables, health stressors, health related hardiness, perception of illness impact, self-perception of health status and psychosocial adjustment to illness; and to determine if perception of illness impact has a direct and/or mediating effect on the relationship between health stressors, health related hardiness, and self-perception of health status and psychosocial adjustment to illness. A cross sectional survey design was used in this study. Sixty individuals of predominantly Asian Pacific Islander ethnicity diagnosed with hemophilia, von Willebrand’s Disease, Factor V or as hemophilia carriers comprised the sample which was drawn from the Hemophilia Treatment Center of Hawaii. All participants were asked to complete five questionnaires: Demographic form and illness information, health related hardiness scale (Pollock, 1990), perception of illness impact scale, self-perception of health status and
psychosocial adjustment to illness scale (Derogatis, 1990). Higher health stressors were associated with higher perception of illness impact, lower perception of health status and poorer psychosocial adjustment to illness. Individuals with higher hardiness were better adjusted to their illness. Higher perception of illness impact was associated with lower self-perception of health status and poorer psychosocial adjustment to illness. Higher self-perception of health status was associated with better psychosocial adjustment to illness. Perception of illness impact did mediate the relationship between health related hardiness and psychosocial adjustment to illness. Perception of illness impact did not mediate the relationship between health stressors and psychosocial adjustment to illness, between health stressors and self-perception of health status, and between health related hardiness and self-perception of health status. The knowledge generated from this study has the potential to impact the existing practices in improving evidence-based nursing practice in caring for individuals with inherited bleeding disorders. Future research is indicated with a large sample to determine differences between diagnosed individuals and carriers, between various Asian Pacific Islander cultural groups, and to determine replicability of the findings from this smaller study sample.
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Chapter 1: Introduction

An individual who is diagnosed with an inherited bleeding disorder is expected to manage his or her condition on a daily basis. This chronic situation can totally disrupt psychosocial functioning and make it more difficult to adjust to the illness. Other researchers have studied this phenomenon in various other chronic illnesses; however, not in individuals with inherited bleeding disorders (Akkasilpa, et al, 2000, Pollack, 1989a, 1989b). Psychosocial problems are not restricted to individuals with one chronic illness and clinically, it is noted that some individuals adjust to chronic diseases better than others. Individuals living with inherited bleeding disorders may also have other chronic illnesses such as hypertension, asthma, diabetes mellitus (DM), congestive heart failure (CHF), arthritis, and hepatitis A, B, C and/or HIV. The aims of this study are to describe health stressors, health related hardiness, perception of illness impact, self perception of health status and psychosocial adjustment to illness in individuals living with an inherited bleeding disorder; to determine relationships between demographic and illness variables, health stressors, health related hardiness, perception of illness impact, self-perception of health status and psychosocial adjustment to illness; and to determine if perception of illness impact has a direct and/or mediating effect on the relationship between health stressors, health related hardiness, and self-perception of health status and psychosocial adjustment to illness. The findings from this study showed that those individuals with higher hardiness were better adjusted to their illness. The knowledge generated from this study has the potential to impact the existing practices in improving evidence-based nursing practice in caring for individuals with inherited bleeding disorders.
Statement of the Problem

Chronic illness refers to an altered health state that will not be cured by a simple surgical procedure or a short course of therapy (Miller, 2000). Chronic illnesses are a considerable health concern throughout the world. Human responses to chronic illness are a challenge for the nursing profession. Many patients with chronic illness have to manage not only one, but also sometimes two or three different chronic illnesses simultaneously. The problem addressed in this proposal pertains to a unique population of individuals with inherited bleeding disorders such as hemophilia, hemophilia carriers, von Willebrand’s Disease (vWD), and other inherited coagulopathies. Many of these individuals are also affected with other illnesses and may be managing more than one chronic illness.

Inherited bleeding disorders are rare disorders affecting a person’s ability to clot adequately. Hemophilia is a sex-linked, genetic disorder characterized by a deficiency or absence of the clotting proteins in plasma. The result is delayed clotting in affected individuals. While deficiencies of any of the clotting factors can occur, factor VIII deficiency (Hemophilia A or Classic Hemophilia) and factor IX deficiency (Hemophilia B or Christmas disease) are the most common deficiencies. Hemophilia has been recognized since biblical times. In the United States, the incidence of hemophilia has been found to be one in 5032 live male births (Souice, et al. 1999). The incidence of Hemophilia A in North America is approximately 1 in 10,000 live male births resulting in approximately 18,000 affected in North America (McDaniel, 2000).

Another common inherited bleeding disorder in females is the hemophilia A or B carrier state in which the factor VIII or IX level may be subnormal and the individual is symptomatic. Although genotypically such a female is a carrier, her factor levels can actually put her in the category of having a “mild hemophilia.” Some studies have concluded that as many as half of all
hemophilia A and B carriers have levels of factor VIII or IX below the normal range and that 20% of carriers have levels below 30%, putting them at risk for excessive bleeding (Nurse’s Guide to Bleeding Disorders, NHF, 2001).

The cause of Von Willebrand Disease (vWD) is a deficiency or defective von Willebrand Factor (vWF). The vWF is an adhesive glycoprotein that has two primary functions. First, the glycoprotein participates in platelet adhesion, spreading aggregation, and second, it binds factor VIII in plasma, protecting it from degradation (Lethagen, 1993). The vWF functions in primary hemostasis by forming an adhesion bridge between platelets and vascular sub endothelial structures and between adjacent platelets at the site of injury. The vWF also plays a role in secondary hemostasis by acting as a carrier protein for factor VIII (Rick, 1994).

Affecting men and women in equal numbers, vWD is the most commonly seen inherited bleeding disorder in women. The exact incidence of vWD is not known; however, it is estimated to affect 1% to 3% of the population worldwide, and about 1% of people in the United States may be affected. Approximately 125 individuals per million (1 in 8000) worldwide have symptomatic vWD. This is roughly twice the prevalence of hemophilia A (Sadler & Gralnick, 1994).

The hepatitis C virus (HCV) infection ranks as the most common chronic blood borne infection in the United States, surpassing human immunodeficiency virus (HIV) (CDC, 1999). HCV infects 1 in 50 Americans and causes 8,000 to 10,000 annual deaths (CDC, 1999). Most of these persons are chronically infected and might not be aware of their infection because they are not clinically ill. Recipients of clotting factor concentrates prepared from plasma pools posed a high risk for HCV infection until procedures to inactivate viruses, including HCV, were introduced during 1985 (Factor VIII) and 1987 (Factor IX). The highest prevalence of infection
is found among those with large or repeated exposure to blood. Persons with hemophilia who are treated with products before inactivation of those products have prevalence rates of HCV infection as high as 90% (Brettler et al, 1990). An estimated 75% of those individuals with hemophilia who are over 12 have chronic HCV infections. Up to one third of these individuals are co-infected with HIV (Eyster, 1991). Very little information is available about HCV infections of contacts of hemophilia patients infected with HCV.

Human Immunodeficiency Virus (HIV) was identified in 1983 as the virus, which causes Acquired Immune Deficiency Syndrome (AIDS). HIV, a retrovirus, attacks and damages the white blood cells of the body. Between 1979 and 1985, 75% of the people treated with plasma products were infected with HIV. More than 50% of severe hemophiliacs experienced seroconversion in early 1983. By 1995, only 30% of those affected had developed AIDS. Nationally there are 2072 individuals affected with HIV and hemophilia (Hemophilia Data Set, 2000).

Based on the information presented above, many individuals living with inherited bleeding disorders may also be dealing with other chronic illnesses such as hypertension, asthma, arthritis, DM, CHF, hepatitis A, B, C and/or, HIV. In combination managing these multiple chronic illnesses can be a stressful and difficult process for these individuals.

There is a lack of research in evaluating health related hardiness in individuals with inherited bleeding disorders. Health related hardiness (HRH) is the personality characteristic that enables individuals to adapt to actual or potential health problems through control, commitment, and challenge (Pollock, 1989a). It is important to determine the relationship between health related hardiness and the health stressors of chronic illness and if health related hardiness is associated with higher psychosocial adjustment to illness and self-perception of health. In Hawaii, this unique population of individuals with inherited bleeding disorders is comprised of a diverse
multicultural group representing Caucasian, Hispanic, Black, Asian, and Pacific Islander. The findings from this study would assist in identifying health disparities among this unique population of individuals, some of who may have multiple chronic illnesses (U.S. Department of Health & Human Services, 1999).

Significance of the Study

The findings from this study have the potential to contribute to a greater understanding of health related hardiness as it relates to a person's self-perception of health and his or her ability to adjust psychosocially to actual and potential health problems associated with inherited bleeding disorders. Knowledge generated from this study may contribute to the development of interventions, thereby assisting these individuals to achieve better adjustment and coping skills in adjusting to their illness(s). By gaining a better understanding of these variables, nurses working with individuals with inherited bleeding disorders can teach and implement health promotion strategies on how to better adjust to and manage his or her bleeding disorder. This expanded insight may also contribute to addressing the theoretical and measurement deficiencies that exist in describing and understanding the construct of health related hardiness and its relationship to health and psychosocial adjustment to illness. Using the PAIS as a screening tool in identifying those individuals who may be mal-adjusted and identifying these individuals early in order to implement interventions.

Purpose of the Study

The aims of this study are to: (1) describe demographic and illness variables, health stressors, health related hardiness, perception of illness impact, self perception of health status and psychosocial adjustment to illness in individuals diagnosed with an inherited bleeding disorder; (2) determine relationships between demographic and illness variables, health stressors, health
related hardiness, perception of illness impact, self perception of health status and psychosocial adjustment to illness; (3) determine if perception of illness impact has a direct and/or mediating effect on the relationships between health stressors, health related hardiness, and self-perception of health status, and psychosocial adjustment to illness.
Chapter 2: Literature Review

Health Stressors

The demands of living with multiple illnesses and the ability to manage those demands can place a considerable amount of stress on these individuals and their families. Some of the illness demands that these individuals and their families encounter are: the tremendous cost of care, stigmatism associated with the disease, insurance issues, fear of unknown viruses, managing drug shortages, negotiating with home care companies, and interacting with the health care system. The estimated annual cost per individual (severe hemophilia) for human plasma factor or recombinant factor products is between $50,000 and $100,000 (NHF, 1998). The psychological, psychosocial, and physiological endurance of managing multiple chronic illnesses is something that individuals living with these chronic illnesses experience. Each chronic illness brings similar as well as unique challenges. The health stressor in this study is defined as any event or situation perceived as threatening to one’s health.

Hardiness

In 1979, Kobasa found that persons who experience high degrees of stress without falling ill have a personality structure different from persons who become ill under stress. This personality characteristic is hardiness (Kobasa, 1979). Hardy persons are considered to possess three general characteristics: the belief that they can control or influence the events of their experience (control), an ability to feel deeply involved in or committed to the activities of their lives (commitment), and the anticipation of change as an exciting challenge to further development (challenge) (Kobasa, 1979, 1981).

Derived from existential personality theory, the hardiness characteristic has been identified as a personality resource that buffers the effect of stress (Pollock & Duffy, 1990). Hardiness was
originally an agricultural concept denoting the quality of crops able to withstand adverse conditions. Subsequently, hardiness was adopted and used by management theorists to examine relationships of personality and stress among workers (Lambert & Lambert, 1987). Hardiness was the first hypothesized, global personality construct found to moderate the stress-illness relationship (Pollock & Duffy, 1990). Other authors broadened the hardiness concept to include such characteristics as a sense of adequacy, authenticity, or self-confidence, among other components (Jennings, 1994).

**Health Related Hardiness**

Pollock (1990) expanded the concept of hardiness to apply specifically to persons with chronic illness. A person with health related hardiness when confronted with a health stressor, possesses the confidence and self-mastery to appraise and modify responses appropriately (control); and cognitively reappraises the health stressor so that it is viewed as stimulating and beneficial or an opportunity for growth (challenge). This in turn leads to motivation and competence in promoting his or her health and coping with the health stressor (commitment) (Pollock, 1990).

Health related hardiness is the personality characteristic that enables individuals to adapt to actual or potential health problems through control, commitment, and challenge (Pollock, 1989a). Pollock (1986) expanded upon the work of Kobasa by examining hardiness in relation to health stress. The purpose of this study was to investigate physiological and psychosocial adaptation of adults with chronic illness; presence of HRH was hypothesized to be positively related to both domains of adaptation. Pollock (1986) initially described the conceptual development and measurement of HRH as a motivating factor in adaptation of adults with chronic illness. The sample included 60 male and female adults with adult onset chronic illness.
(arthritis n = 20, diabetes n = 20, hypertension n = 20) for at least one year. The study investigated factors that promoted physiological and psychosocial adaptation in chronically ill adults. The hypothesis that the presence of the hardiness characteristic was significantly correlated with physiological (r = .43, p < .05) and psychosocial (r = .62, p < .01) adaptation was supported for subjects with insulin dependent diabetes, but not for the subjects with hypertension or rheumatoid arthritis. The findings were reported as suggesting that specific psychosocial activities may be facilitated by the presence of health related hardiness, thus supporting the indirect effect of hardiness on adaptation. The indirect effects were noted as influenced role function, social support and intrapsychic functioning.

Pollock (1989b) investigated the relationship between the physiological adaptive responses of adults with insulin dependent diabetes mellitus (IDDM), and stress appraisals, coping patterns, health-related hardiness, and sociodemographic variables. A descriptive research method was used and the typical subject (N = 30) was a middle-aged white female who worked outside the home and had IDDM for less than 10 years. This study supports the thesis that how one perceives one’s situation (stress appraisals) and what one does about the situation (coping strategies) are important variables in predicting adaptation to a chronic illness. Emotion-focused coping was significantly correlated with the appraisal of threat and challenge (r = .61, p < .001). Participants who perceived their illness as a threat or challenge were more likely to use emotion-focused coping strategies. There was no significant correlation between the outcome-appraisal categories of harm and benefit and coping patterns. A significant negative correlation was found between appraisals of harm and benefit with total health related hardiness (r = -.49, p < .05). Persons with the health related hardiness characteristic perceived their illness as a potential harmful or beneficial factor rather than a threat or challenge event. There were no significant
correlations between the anticipatory appraisals and the total health related hardiness score. A significant negative relationship was found between mixed-focus coping and total hardiness \((r = -.44, p < .05)\). Significant negative correlations were found between emotion-focused coping strategies and hardiness \((r = -.44, p < .05)\). There were no significant relationships between hardiness and problem-focused coping. Significant correlations were also found between physiological adaptation and total hardiness scores \((r = -.42, p < .05)\) and the sub-concept of commitment \((r = -.45, p < .05)\). Multicollinearity was not found among the predictor variables.

The presence of health related hardiness characteristic was related to better physiological adaptation to chronic illness. Health related hardiness also was related to how one perceives the situation and what one does about the situation. A multiple regression analysis was used to determine which variables were predictive of psychosocial adaptation in adults with IDDM 56% of the variance in predicting physiological adaptation was accounted for by five variables: outcome-appraisal 17%, Mixed-coping pattern 14%, hardiness 11%, patient education 9% and emotion focused coping 5%.

Pollock, Christian, & Sands (1990) examined whether adaptive responses of adults with various chronic illnesses were significantly different. A sample of \((N = 211)\) participants in three chronic illness groups were included: multiple sclerosis \((n = 44)\), hypertension \((n = 42)\) and rheumatoid arthritis \((n = 125)\). Physiologic adaptation was measured by one of three scales according to diagnosis. Physiologic adaptation was significantly different among the three chronic illness groups; however, psychological adaptation did not differ among the groups. This suggests that although each chronic illness has disease-specific physiologic changes, the nature of the psychological adaptation process is similar. The health related hardiness characteristic was the only major variable that was related to both physiologic and psychosocial adaptation.
Pollack & Duffy (1990) described the development and psychometric evaluation of the HRHS (Health Related Hardiness Scale). It was hypothesized that health related hardiness consisted of three discrete but related dimensions (control, commitment and challenge). Control was defined as the sense of mastery or self-confidence needed to appraise and interpret health stressors appropriately. Committed persons are those who tend to become involved in, rather than alienated from, various activities in life. Challenge refers to the anticipation of change as an exciting stimulus rather than a threat. Only two dimensions were supported by the principle components analyses and reliability estimates. The first factor (20 items) encompassed the dimensions of challenge and commitment, and the second factor (14 items) accounted for the control dimension. The HRHS has three main advantages over Kobasa’s hardiness measure: health specificity, measurement of the presence of factors (commitment/challenge and control) rather than the absence to determine hardiness and an easy scoring method.

Pollock (1993) summarized the results of a program of nursing research, which included identifying predictors of adaptation to chronic illness and determining whether adaptive responses differed by diagnostic groups. This study utilized a cross-sectional design with a retrospective sample of (N = 597) adults who had insulin dependent diabetes mellitus (n = 251); multiple sclerosis (n = 137); hypertension (n = 113) and rheumatoid arthritis, (n = 96). The health related hardiness characteristic, ability to tolerate stress, and health promotion activities were related positively to both physiological adaptation and psychosocial adaptation for the total sample. The findings from this study support the direct effects of hardiness on adaptation as well as the indirect effects through health promotion activities. Hardy persons were more likely to engage in health-related activities and these activities were related to better physiological adaptation.
Summary of Pollock's Work

A health related hardiness concept was proposed to link hardiness more closely to the context of health and illness (Pollock, 1986). Pollock (1986) extended the hardiness construct to study adaptation in chronic illness. The synthesis of the health related hardiness characteristics on adaptation to actual or potential health problems incorporate concepts from existential psychology, coping and adaptation, and development tasks of adulthood (Pollock, 1989a). Pollock (1986) found control and commitment of health related hardiness significantly related to higher psychosocial adaptation in individuals with diabetes mellitus, commitment was significantly related to psychosocial adaptation in individuals with hypertension and rheumatoid arthritis. This was supported by the perception of the subjects with diabetes who believed that they had some control over their disease, whereas the individuals with rheumatoid arthritis felt helpless in controlling their disease even if their therapy was effective (Pollock, 1986).

Pearson’s correlation for the total group revealed significant relationships between hardiness and psychosocial adaptation, (r = .42, p = .01).

Other Research Studies Using HRHS

Other researchers have also studied health related hardiness in various health situations using Pollock's health related hardiness instrument. Newtown (1999) investigated the relationship between health related hardiness and sense of coherence (defined as a pervasive, enduring confidence that 1) the stimuli deriving from one’s internal and external environment in the course of living are structured, predictable, and explicable; 2) the resources are available to one to meet the demands posed by these stimuli; and 3 these demands are challenges, worthy of investment and engagement) in post liver transplant patients’ return to work. The study was a retrospective cross-sectional design and a convenience sample was used. The subjects (N = 230).
were liver transplant patients from a large Midwestern liver transplant program who were all older than 18 years of age. The mean scores for hardiness and sense of coherence were higher for working than for nonworking recipients. Length of time after transplant, had a statistically significant effect on recipient return to work \( (p = 0.000) \). The time frame producing the largest number of workers (73%) was 5 years or more after transplant; only 29% of the recipients were working less than 1 year after transplant. A Pearson's product moment correlation coefficient indicated that hardiness had a positive relationship with sense of coherence \( (r = 0.46) \). The findings from this study indicated that sense of coherence and hardiness each had a direct and independent positive effect on a post-liver transplant patient's return to work. The results of this study also acknowledge that sense of coherence, independent of hardiness, has strong predictive capabilities for post-liver transplant return to work. The major limitation of this study was the sampling technique.

In a cross sectional study, Akkasilpa, et al (2000) investigated the relationship between two specific psychological constructs (health related hardiness and uncertainty) with fibromyalgia (FM) tender points (TP) in 173 mostly female (94.2%) patients with systemic lupus erythematosus (SLE). The HRHS and the Mishel Uncertainty in Illness Scale (MUIS) were used in the study. No significant association was found between FM TP and age or the level of education. There were significant associations between the HRHS scores and the number of FM TP \( (p = 0.0108) \) and the level of education \( (p = 0.0012) \), but not with age, sex, or race. There were also significant associations between the scores of the MUIS and FM TP \( (p = 0.0001) \) and race \( (p = 0.007) \), but not with age, sex, or the level of education. SLE patients with FM TP were less likely to be good copers (Akkasilpa et al., 2000).
Schott-Baer et al (1995) used the HRHS in a descriptive correlational study to clarify some of the relationships found in the earlier investigation by including health-related hardiness as a possible mediating factor between a caregiver's self care agency (defined as a person's ability to engage in self-care) and the negative impact of the care giving experience. This study used a convenience sample of N = 53. They were 39 women and 14 men. A significant negative correlation was found between objective burden and dependent care. A significant positive correlation was found between self-care agency scores and the commitment/challenge and control subscales of health related hardiness scores (Schott-Baer et al., 1995).

An intervention study by Webster & Austin (1999) evaluated a clinical nursing intervention, the Wellness Program, to determine whether this program fostered the development of thoughts, feelings and behaviors associated with health related hardiness. The study design was a quasi-experimental nonequivalent control group. This study was a partial replication of a descriptive correlation study that examined the relationship between dependent care, caregiver burden, and health related hardiness and self-care agency of caregivers. The subjects were adults (N = 10) who believed their lives were being affected by stress. Their ages ranged between 23 and 55. Findings from this study suggest that individual changes in levels of commitment/challenge and control are possible with a brief psycho-educational program. Subjects in the treatment group demonstrated cognitive transformation of life events as they experienced a statistically significant decrease in their average level of distress. Limitations to the study were the small sample size and lack of randomization of participants (Webster & Austin, 1999).

Narsavage and Weaver (1994) used the HRHS to examine the relationship between physiologic status, coping and health related-hardiness and exercise and functional status. The subjects (n = 96) were outpatients with chronic obstructive pulmonary disease (COPD)
diagnosed an average of 11.61 years. The majority of participants were men (n = 85). Age of subjects ranged from 40 to 84 years with a mean age of 65.5 years. The distance walked in 5 minutes measured exercise ability. Pulmonary Function Tests were used to measure physiological status. Physiologic status was correlated with the outcome variable of exercise ability (r = .37, p < .01) and functional status (r = .29, p < .01). Problem-solving coping significantly correlated with exercise ability (r = .23, p < .05). The commitment component of hardiness was related to both exercise ability and functional status (r = -.23, p < .05). The challenge component (r = -.21), as well as total hardiness, were related to exercise ability (r = -.20, p < .05), but not to functional status. The findings from this study suggest that the hardier COPD patients who were characterized by commitment and challenge used more problem-solving techniques and had improved exercise ability.

Ross (1991) examined the relationship between health related hardiness and self-management compliance. The purpose of this study was to determine if the presence of the health related hardiness personality characteristic influences compliance. This was a correlational design with a sample of (N = 50) elderly patients with diabetes. Age of subjects ranged from 65 to 80 years old. Participants were primarily male, white, married and retired. Results indicated a significant relationship between the total HRHS and compliance (r = -.60, p < .05). No significant correlations were found between the three subscales of the HRHS and compliance. Stepwise multiple regression analyses for the dependent variable of compliance indicated that three variables (total health related hardiness scale, hospitalization for nondiabetic illness and age) accounted for 49% of the variance in predicting compliance. Findings suggest that the degree of health related hardiness in individuals with diabetes might predict compliance to a prescribed diabetic regimen.
Nicholas (1993) examined the relationship among health related hardiness, self-care practices and perceived health status in older adults. A random sample of older adults over 55 years of age (N = 72) who were residents of a small northwestern town in the United States was recruited. A Visual Analogue Scale and the Personal Lifestyle Questionnaire measured the extent to which individuals engaged in health promotion or self-care activities. An illness index was developed by the researcher to identify type and number of illnesses. A source of multicollinearity was found between health related hardiness and self-care practices (r = -0.68, n = 64, p < 0.001) with 46% shared variance between health related hardiness and self-care practices. Health related hardiness was significantly related to perceived health status (r = -0.68, n = 66, p < 0.001). Health related hardiness and self-care practices were significant predictors of perceived health status in older adults.

Nicholas and Leuner (1999) examined the relationship between health related hardiness, social support, health status and selected sociodemographic variables, including ethnicity, in an older population. This was a descriptive study with a sample size of (n = 110) participants whose age ranged from 55 to 93. There were 33 men and 77 women. Nearly 44% (n = 48) were married. Older African-American adults represented 27% of the sample (n = 30) and older Anglo-American adults 73% of the sample (n = 80). Personal Resource Questionnaire (PRQ85-Part 2) was used to measure perceived social support and a Visual Analogue Scale was used to measure perceived health status. A significant relationship was found between hardiness and social support (r = 0.41, p < 0.01) and hardiness and perceived health status (r = 0.78, p < 0.001). A significant positive relationship was found between social support and perceived health status. The findings of this study indicated that health related hardiness and social support were
significantly related to perceived health status in older African-American and Anglo-American adults.

Cross Cultural Research and Health Related Hardiness

Wang (1999) verified the feasibility of the HRHS cross-culturally and sought to compare the effects of health related hardiness between nurses in the United States and Taiwan. This was a cross-sectional and cross-cultural study that used a convenience sample of \( N = 778 \); \( n = 163 \) from the United States, 615 from Taiwan. The HRHS was translated from English to Mandarin Chinese for this study. A bilingual Chinese faculty member then translated the Chinese version back into English. The findings from this study verify that Pollock’s HRHS is valid for measuring the health related hardiness construct as exemplified by the strong factor loading obtained from this study, which ranged from 0.43 to 0.68 for American nurses and 0.47 to 0.68 for Taiwanese nurses. Both factors are identical from Pollock’s previously reported factor loading (0.41 to 0.68). This study is significant in verifying the validity of the HRHS as a useful tool in non-English-speaking cultural groups. The HRHS is a valid and reliable tool to measure health related hardiness cross-culturally.

Martin et al. (1999) looked to identify and quantify associations between HRH and function, self-assessed health morbidity, health behaviors and selected demographic variables that are predictive of HRH in urban older African American women who have chronic illness. A correlational design and a probability sample of every fourth older African American woman scheduled for an appointment in a medical clinic was used. Subjects were \( n = 100 \) African American women, age 60 years or older, diagnosis of hypertension (90%), diabetes mellitus (41%) and/or osteoarthritis (74%), and ability to respond in English. The HRHS was used to measure health related hardiness, and the Sickness Impact Profile (SIP) was used to measure
quality of life. Two variables were found to have significant relationships with HRH: years of education \( (r = 0.39) \) and the SIP score \( (r = 0.27) \). With health related hardiness as the dependent variable, years of education and the SIP score explained 20% of the variance in health related hardiness, accounting for 15% \( (p < 0.0001) \) and 5% \( (p < 0.02) \) of variance respectively. Issues regarding the validity of HRH prediction models and the cultural appropriateness of current methods of assessing HRH in older African-American women (Martin et al., 1999) were addressed. Prediction models developed with samples of predominately young, well-educated, Anglo-American men and women of middle socioeconomic status are invalid for southern, older African-American women of low socioeconomic status with chronic illnesses. Generalizations about the expression and determinants of HRH in chronically ill adults may be invalid for older African-American women.

Pollock (1999) summarizes health related hardiness research completed with adults that has relevance for ethnic populations, describes programs of translation of the HRHS and discusses recommendations for future work. The HRHS has been used in more than 250 studies that focused on either health promotion or adaptation to health problems. The major limitation in using the HRHS with different ethnic populations was that the subjects needed to read English or understand spoken English.

Velasco-Whetsell and Pollock (1999) described the development and initial evaluation of a Spanish language version of the HRHS and reported on the Spanish language HRHS. The second study back translated the Spanish Health Related Hardiness Scale (SHRHS), which demonstrated a strong correlation with the first translation obtained by Boytell, Velasco-Whetsell, and Coffin (1996) further supporting the accuracy and reliability of the SHRHS. Results, with regard to
translation and equivalence, further support the accuracy of the translation and its equivalence to the English version.

**Psychosocial Adaptation to Chronic Illness**

Several studies have been reported on psychosocial adaptation to chronic illnesses; however, no studies have been reported on psychosocial adaptation and inherited bleeding disorders. The primary focus of five research studies investigated by Pollock (1986, 1989) and colleagues (Pollock, Christian & Sands, 1990; Pollock & Sands, 1992) was the development of knowledge about adaptive responses to chronic illnesses. Psychosocial adaptation did not differ among the chronic illness groups, whereas there were significant differences in physiological changes. This implies that the process of psychosocial adaptation is similar across different chronic diseases (Pollock, 1993).

**Self-Perception of Health**

Self-perception of health is defined in this study as an individual’s understanding of his or her current physical condition. Maddox and Douglas (1973) have concluded that self-ratings of health clearly have utility as a measure in research. In a study by Nicholas and Leuner (1999) a significant relationship was found between hardiness and perceived health status ($r = 0.78$, $p < 0.001$). The relationship between social support and perceived health status was also supported ($r = 0.66$, $p < 0.001$), with a significant positive relationship found between the two variables. Nicholas (1993) found age to be significantly related to perceived health status ($r = -0.31$, $n = 72$, $p < 0.004$). The total sample of 72 included 39 women (54.2%) and 33 men (45.8%). The elderly subjects ages ranged from 55 to 92 years. The mean age was 69.5 years (SD = 7.3). Older persons have better perceived health status.
Hardiness was also significantly related to perceived health status ($r = -0.68$, $n = 66$, $p < 0.001$), with individuals who were high on the hardiness characteristic indicating higher perceived health status scores. A moderately strong correlation was also found between self-care practices and perceived health status ($r = 0.46$, $n = 70$, $p < 0.001$). Hardiness and self-care practices were also found to have a statistically additive effect on perceived health status scores.

In summary, Pollock (1989a) found health related hardiness significantly related to higher levels of perceived health status. Hardiness and social support were also significantly related to perceived health status in older adults (Nicholas, 1999). Furthermore, hardiness and self care practices were also found to be important predictors of perceived health status in older adults (Nicholas, 1993).

**Pollock’s Model**

Pollock’s Model has identified three contextual stimuli (health promotion activities, ability to tolerate stress and health related hardiness) that influence both physiological and psychosocial adaptation to chronic illness. An individual’s perception of the impact on adaptation is consistent with Roy’s thesis that the regulator (a major coping process involving the neural, chemical and endocrine system) and cognator (a major coping process involving four cognitive-emotional channels: perceptual and information processing, learning, judgment and emotion) control processes act through the four-effector modes (physiological, self-concept, role function and interdependence) to promote physiological and psychosocial integrity as depicted in Pollock’s model (Roy, 1984). Results from this study provide support for the relationship posited by the theoretical framework depicted in Pollock’s model. The perception of the disability caused by the illness is an important variable in the process of adapting to a chronic illness (Pollock, 1993). Other researchers have concurred that the best index of psychological
response to chronic illness is the individual’s perception of the disability produced (Westbrook & Viney, 1982).

*Figure 1.*

**Pollack’s Model**

![Pollack's Model Diagram]

**Study Model**

The study model was developed after a review of the literature and consideration of the unique strengths and limitations of the population with inherited bleeding disorders. Health related hardiness has a direct relationship with health stressors. Both health stressors and health related hardiness are depicted as having both direct relationships to psychosocial adaptation (psychosocial adjustment to illness and self-perception to health status) to illness as well as an indirect relationship via perception of illness impact. Perception of illness impact was chosen as a potential mediating variable between psychosocial adaptation (psychosocial adjustment to illness and self-perception of health status), health stressors and health related hardiness. Perception of illness impact is depicted as having a direct relationship to psychosocial adaptation. The mediator represents a process or mechanism, often intrinsic to an individual that
accounts for the relationship between a predictor variable and an outcome variable. The mediator specifies how or why the relationship occurs (Baron & Kenny, 1986). In previous studies (Nicholas, 1993 & Pollock, 1986) perception of illness impact has been shown to be a significant variable as it is the individual’s subjective appraisal of how disabling a chronic illness can be.

Figure 2.

*Study Model I.*

![Study Model I Diagram]

Figure 3.

*Study Model II.*

![Study Model II Diagram]
Research Questions

1) What is the relationship between demographic and illness variables, health stressors, health related hardiness, perception of illness impact, self-perception of health status and psychosocial adjustment to illness in individuals with inherited bleeding disorders?

2) Does perception of illness impact mediate the relationship between health stressors and psychosocial adjustment to illness?

3) Does perception of illness impact mediate the relationship between health related hardiness and psychosocial adjustment to illness?

4) Does perception of illness impact mediate the relationship between health stressors and self-perception of health status?

5) Does perception of illness impact mediate the relationship between health related hardiness and self-perception of health status?

Hypotheses

1) Health stressors will be positively associated with psychosocial adjustment to illness* (lower psychosocial adjustment).

2) Health stressors will be negatively associated with self-perception of health status.

3) Health related hardiness will be negatively associated with psychosocial adjustment to illness* (higher psychosocial adjustment).

4) Health related hardiness will be positively associated with self-perception of health status.

5) Perception of illness impact will be positively associated with psychosocial adjustment to illness* (lower psychosocial adjustment).
6) Perception of illness impact will be a negatively associated with self-perception of health status.

* Note: Lower scores on instrument used to measure psychosocial adjustment to illness (PAIS-SR) indicated higher psychosocial adjustment to illness.

7) Perception of illness impact will mediate the relationship between health stressors and psychosocial adjustment to illness.

8) Perception of illness impact will mediate the relationship between health related hardiness and psychosocial adjustment to illness.

9) Perception of illness impact will mediate the relationship between health stressors and self-perception of health status.

10) Perception of illness impact will mediate the relationship between health related hardiness and self-perception of health status.
Chapter 3: Methodology

*Research Design and Setting*

A cross sectional method design (descriptive and predictive) was used in this study. Recruitment of participants was carried out at the Hemophilia and Thrombosis Treatment Center of Hawaii. This outpatient setting is a federally funded clinic located within Kapiolani Medical Center for Women and Children. Kapiolani Medical Center is one of the Pacific region’s leaders in the care of women and children, with 232 beds and 90 bassinets. As the Regional Perinatal Center of the Pacific, this institution annually conducts more than $2 million in research activity, and is a major teaching hospital affiliated with the University of Hawaii’s John A. Burns School of Medicine.

*Population and Sample*

The Hemophilia and Thrombosis Treatment Center of Hawaii (HTTCH) had an active population of 158 in 2002, a 21% increase from 2001. The portion with Hemophilia A is 52% and Hemophilia B is 15%, 23% had vWD and 11% had other inherited coagulopathies. There are 5 known patients with HIV and hemophilia treated at the HTTCH (Hemophilia Data Set, 2002). The ethnic groups who have hemophilia A, hemophilia B, vWD, and/or other genetically transmitted bleeding disorder are: Asian/Pacific Islander, 51%; White, 22%; African-American, <1%; and other ethnicities (Mixed Race, Native American and other), 26% (Hemophilia Data Set, 2002). The number of vWD rose from 23% to 28% in 2002. There has been a steady increase in the number of vWD patients being followed at the HTTCH. The establishment of the HTTCH in 1998 at Kapiolani Medical Center may attribute to the increased number of patients identified and followed at the center. This increase may also be attributed to better referral and diagnosis of these patients.
A convenience sample was recruited and consisted of adults 18 years and older with inherited bleeding disorders who may also be diagnosed with one or more chronic illnesses: [Hemophilia (A or B), Hemophilia Carriers (A or B), Vwd] as well as possibly hepatitis A, hepatitis B, hepatitis C, HIV or other chronic illnesses such as hypertension, asthma, diabetes mellitus, arthritis, and congestive heart failure. Eligibility criteria were: Males and females with inherited bleeding disorders who were 18 years or older, able to speak and read English, and who received care from a comprehensive clinic at the federally funded hemophilia treatment center in Hawaii. Exclusion criteria: Cognitive or neurological impairments.

Power Analysis

A power analysis was estimated utilizing the G-Power, a general power analysis program (Erdfelder, Faul, & Buchner, 1996). Based on a medium effect size (0.15), an alpha of 0.05, and a power of 0.80, for two predictors (health stressors and perception of illness impact; health related hardiness and perception of illness impact respectively) the sample size for 2-tail correlations would be 64 and the sample size for F-tests multiple regression would be 68.

Instruments

All participants were asked to complete five questionnaires: Demographic form and illness information, Health Related Hardiness Scale (HRHS), Perception of Illness Impact Scale (PIIS), Self- Perception of Health Status (SPHS) and Psychosocial Adjustment to Illness Scale (PAIS-SR). A description of these measures and how they were used in statistical analysis follows.

Demographic variables collected and measured at entry to the study were age, gender, ethnicity, marital status, education, employment, occupation, income, number of children, and living situation. Self-reported illness information collected included type of bleeding disorder
and date of diagnosis and the presence of other chronic illnesses such as hepatitis A, B, C, hypertension, asthma, diabetes mellitus, congestive heart failure, and arthritis (See Appendix A).

**Health Stressors**

In this study, the health stressor was the presence of an inherited bleeding disorder and any other chronic illnesses. Additionally, the concept of health stressor was introduced to assist in understanding health related research. A health stressor is defined as any event or situation perceived as threatening to one’s health (Pollack, 1990). In this study, the health stressor is the presence of an inherited bleeding disorder and any other chronic illnesses. A cumulative or summed score will be used in evaluating the health stressors. Each item was assigned a score of 1 for each chronic illness the participant has and a total score was derived. The maximum score varied depending on how many chronic illnesses the participant had.

**Health Related Hardiness**

The HRHS was first developed by Pollock in 1986. The HRHS was developed to measure the hardiness construct in health related hardiness (HRH). Since its inception, the instrument has been used in a variety of health related studies with adults with chronic illness, including health promotion research (Pollock, 1999).

The HRHS was developed to more adequately measure the hardiness characteristic in chronically ill individuals. Possession of the characteristics of control, commitment and challenge in relation to health may account for successful resistance to the negative effects of stress. The HRHS has been shown to be a valid and reliable instrument in health-related hardiness research. Of particular importance is the fact that the domain-specific subscales of the HRHS showed higher predictive validity than Kobasa’s hardiness measure (Pollock, 1989a).
The HRHS was developed using the theoretical definitions of health related control, commitment and challenge. The health related hardiness framework states that when a hardy person is confronted with a health stressor, he or she possess the confidence and self-mastery to appraise and modify responses appropriately (control) and cognitively reappraise the health stressor so that it is viewed as stimulating and beneficial or an opportunity for growth (challenge) which in turn is evidenced by motivation and competence in promoting his or her health and coping with the health stressor (commitment), (Pollack, 1990).

The first version of the HRHS consisted of 48 items measured on a 6-point Likert scale (strongly agree to strongly disagree). A pilot study of the questionnaire was pretested for readability, clarity, meaning and response variance on a sample of 53 graduate nursing students. Based on their evaluations, several items were clarified and three new items were added for a total of 51 items. A readability index was computed, indicating that the HRHS required at least an eighth grade reading level. Frequency distributions indicated that respondents chose the full range of responses for most items (Pollack & Duffy 1990).

Following the pretest, a panel of three experts who examined each item for congruence with the health-related hardiness definitions of control, commitment and challenge evaluated the 51-item HRHS. This first version was lengthy to administer and difficult to score (Pollack & Duffy 1990).

Content validity of the HRHS was addressed by asking a panel of judges, faculty and doctoral students with clinical experience in adult health to evaluate the representativeness of the control, commitment, and challenge items on the basis of the operational definitions. Agreement among the judges concerning the items that best measured the control, commitment and challenge dimensions of hardness was 0.85. Several versions of the HRHS have been developed
and it is difficult to ascertain which version of the HRHS is being used in specific published reports. Several studies used the HRHS with 51, 48 and 40 items (Narvargase, 1994, Nicholas, 1993, Pollock, 1989a, Pollock & Duffy, 1990, & Ross 1991). Other studies that used the 34 item HRHS (Akalipsisa, 2000, Martin, 1999, Newtown, 1999, Nicholas, 1999, Schott, 1995, & Wang, 1999).

Table 1.

*Development of the HRHS.*

<table>
<thead>
<tr>
<th>HRHS Description</th>
<th># Items</th>
<th>Sample</th>
<th>Description of items</th>
<th>Chronbach's Alpha</th>
<th>Reliability (Chronbach's Alpha)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Initial Comparison between Kobasa Hardiness Scale &amp; Pollock HRHS</td>
<td>Version 1 (Pollock, 1986)</td>
<td>48</td>
<td>n = 50</td>
<td>Commitment (15 items), Challenge (15 items) &amp; Control (18 items).</td>
<td>.82 for control, .74 for commitment and .84 for challenge.</td>
</tr>
<tr>
<td></td>
<td>Version 1a (Pollock, 1986)</td>
<td>48</td>
<td>n = 60</td>
<td>Commitment (15 items), Challenge (15 items) &amp; Control (18 items).</td>
<td>Alphas for control. 84, commitment .78 and challenge .82</td>
</tr>
<tr>
<td></td>
<td></td>
<td>48</td>
<td>n =30</td>
<td>Commitment (15 items), Challenge (15 items) &amp; Control (18 items).</td>
<td>.84 control, .82 commitment, and .74 challenge</td>
</tr>
<tr>
<td></td>
<td>Version 2.1</td>
<td>42</td>
<td>n = 65 (Pilot study)</td>
<td>Control (18 items), commitment (12 items) and challenge (12 items)</td>
<td>Control .82, Commitment .74 and Challenge .65</td>
</tr>
<tr>
<td></td>
<td>Version 2.2 (Narsavage &amp; Weaver 1994)</td>
<td>40</td>
<td>n = 96</td>
<td>Study does not give the breakdown of the different subscale items.</td>
<td>.72 control, .72 commitment and .64 challenge.</td>
</tr>
<tr>
<td>(Ross, 1991)</td>
<td>40</td>
<td>n = 50</td>
<td>Control 14 items, Commitment 13 items, 13 items challenge</td>
<td>.59 for control, .61 for commitment and .49 for challenge</td>
<td>.74</td>
</tr>
<tr>
<td>(Nicholas, 1993)</td>
<td>40</td>
<td>n = 110</td>
<td>Control 14 items, Commitment 13 items and Challenge 13 items.</td>
<td>78 for control, .82 for commitment and .76 for challenge</td>
<td>.86</td>
</tr>
<tr>
<td>Version 3.1 (Pollock, et. al 1990)</td>
<td>51</td>
<td>n = 211</td>
<td>Control (21 items), Commitment (15 items), &amp; Challenge (15 items).</td>
<td>Re-test was .90 for 2 weeks and .80 for 3 months) In this study, .84 for control, .71 for commitment and .80 for challenge.</td>
<td>.89</td>
</tr>
<tr>
<td>Version 3.2 (Wang, 1999)</td>
<td>34</td>
<td>n = 778</td>
<td>Control (14 items), Commitment (7 items) &amp; challenge (13 items).</td>
<td>0.87 for Control, Commitment .74 and Challenge .81.</td>
<td>.91</td>
</tr>
<tr>
<td>Current (Schott-Baer et al. 1995)</td>
<td>34</td>
<td>n = 53</td>
<td>Commitment/challenge subscale (20 items) and 14 on the control subscale</td>
<td>.87 for the control</td>
<td>.91</td>
</tr>
<tr>
<td>Current (Martin, 1999)</td>
<td>34</td>
<td>n = 100</td>
<td>Commitment/Challenge (20 items) or control (14 items).</td>
<td>Test-retest reliability at 6 months was 0.76 for the total scale and for the control and .78 for Commitment/challenge.</td>
<td>.88</td>
</tr>
</tbody>
</table>
Depending on the purpose of the investigation, the HRHS can be used to measure the unitary construct of health-related hardiness and/or the two dimensions of commitment/challenge and control. Results of a principal component analysis with chronically ill subjects (n = 389) supported the two factors rather than three (Pollock & Duffy, 1990). The first factor (20 items) encompassed commitment and challenge while the second factor (14 items) accounted for the control. The two factors explained 32.1% of the variance (Pollock & Duffy, 1990).

Pollock (1990) addressed convergent validity. The Kobasa Hardiness Inventory and the HRHS were both administered to healthy adults (N = 50). A correlation of .54 between the two scales supported the idea that the HRHS was measuring hardiness but was also sufficiently different from Kobasa’s scale. The HRHS, but not Kobasa’s Hardiness Scale, was positively correlated with perceived health status, engagement in health promotion activities and use of resources. A significant but moderate correlation was found between hardiness and physiologic adaptation. Presence of the hardiness characteristic was also related to how one perceives chronic illness and what one does about the situation.

Commitment and challenge items loading together suggest that they are closely related and not discrete dimensions in a health specific context. In other words, commitment to adjusting to a health stressor such as chronic illness is also a challenge. Persons with HRH are challenged rather than threatened when confronted with a health stressor, this challenge then becomes a
personal commitment. Hardy individuals dealing with a chronic health problem may not separate health into discrete categories but appraise the condition as a challenge because they are committed to maintain their health (Pollock, 1990).

Internal reliabilities are .91 and .87 for the 20-item commitment/challenge subscale and the 14-item control scale respectively. Test-retest reliability (N = 150) at six months, was .76 for the total scale, .74 for the challenge/commitment subscale and, .78 for the control subscale (Pollock, 1990).

In this study, the most current version of the HRHS will be used which contains 34-items measuring two subscales: control 14-items, commitment/challenge 20-items (See Appendix B). The instrument is now constructed to measure commitment and challenge as one subscale. Scores for the total HRHS range from 34 to 204 with higher scores indicating more health related hardiness. Item scores will be summed to obtain an overall HRHS score that was used for analysis.

Perception of Illness Impact

The variable of perception of illness impact was measured using a 10-point Likert scale. Instructions asked the participant to circle the number that corresponded to the amount their bleeding condition had affected their ability to do daily activities (work, school) in the past twelve months from not affected at all to affected a great deal with a possible range of 1-10 with endpoints of not affected at all to affected a great deal. Higher scores indicated poorer perception of illness impact (See Appendix C).

Self-Perception of Health Status

The variable of self-perception of health was measured using a four point Likert scale that provided choices from poor to excellent. This measure was used because it assesses the
individual’s total multifaceted view of health. The instrument has strong reliability in self-assessment of well-being and correlates strongly with clinician ratings (Nicholas, 1999). Higher scores will indicate better self-perception of health (See Appendix D).

*Psychosocial Adjustment to Illness*

The Psychosocial Adjustment to Illness Scale (PAIS) (Deragotis, 1986) is a 46-item, multiple domain, semi-structured questionnaire designed to quantify and assess the quality of a patient’s psychosocial adjustment to a current medical illness or the sequelae of a previous illness. The PAIS – SR (Self Report) is a self-report version of the scale. The PAIS-SR scale is composed of seven primary domains of adjustment: I. Health Care Orientation (8 items), II. Vocational Environment (6 items), III. Domestic Environment (8 items), IV. Sexual Relationships (6 items), V. Extended Family Relationships (5 items), VI. Social Environment (6 items) and VII. Psychological Distress (7 items). Each PAIS-SR item is rated on a 4-point (0 to 3) scale of adjustment, with higher ratings indicating poorer adjustment status. On the PAIS-SR, scale direction is alternated on every other item (2, 4, and 6) to help reduce position response biases (Derogatis, 1986).

The instrument can be administered either as an interview or as a self-report questionnaire and can be completed in 20-25 minutes. In this study, the self-report questionnaire was used. The reference point for the administration of most questions is a reflection on the most recent 30 days in the respondent’s life (Derogatis, 1986)

The original work for the development of the PAIS was on patients suffering chronic, life-threatening illnesses, particularly cancer. Subjects for the reliability study were 37 patients with Hodgkin’s disease. In addition, a validation study was completed which included an independent sample of 38 parents whose children had received treatment for Hodgkin’s disease or solid
tumors. Disease specific norms are available for some chronic illness groups; however, not for individuals with inherited bleeding disorders. Reliability Coefficients are available for three distinct normative samples: 269 renal dialysis patients, 89 lung cancer patients, and 69 cardiac patients.

Table 2.

*Reliability Coefficients.*

<table>
<thead>
<tr>
<th>PAIS DOMAIN</th>
<th>Renal Dialysis N=269</th>
<th>Lung Cancer N=89</th>
<th>Cardiac N=69</th>
</tr>
</thead>
<tbody>
<tr>
<td>I. Health Care Orientation (HCO)</td>
<td>.63</td>
<td>.83</td>
<td>.47</td>
</tr>
<tr>
<td>II. Vocational Environment (VE)</td>
<td>.81</td>
<td>.87</td>
<td>.76</td>
</tr>
<tr>
<td>III. Domestic Environment (DE)</td>
<td>.67</td>
<td>.68</td>
<td>.77</td>
</tr>
<tr>
<td>IV. Sexual Relationships (SR)</td>
<td>.80</td>
<td>.93</td>
<td>.83</td>
</tr>
<tr>
<td>V. Extended Family Relationships (EFR)</td>
<td>.66</td>
<td>.12</td>
<td>.62</td>
</tr>
<tr>
<td>VI. Social Environment (SE)</td>
<td>.78</td>
<td>.93</td>
<td>.80</td>
</tr>
<tr>
<td>VII Psychological Distress (PD)</td>
<td>.80</td>
<td>.81</td>
<td>.85</td>
</tr>
</tbody>
</table>
In three published studies internal reliability estimates (coefficients) for the domain scores of the PAIS-SR were: .63 - .80; .68 - .93; and .47 - .85. Validation of the PAIS has been extensive, with studies supporting convergent, predictive and construct validity as well as confirmation of dimensional structure (Derogatis & Derogatis, 1986, Derogatis & Fleming, 1996).

Construct validity: A principal components analysis was accomplished with subsequent rotation to a normalized varimax solution (factors are rotated for the best fit and the factors are uncorrelated). Seven substantive dimensions, as hypothesized, were identified, accounting for 63% of the matrix variance. These seven domains were subjected to analysis and found to have low inter-domain correlation, but high correlation with the PAIS Total Score (Derogatis, 1986). In the varimax solution, Factor I accounted for 18% of the variance, with the remaining dimensions being associated with 10, 9, 8, 7, 7 and 5% respectively. (Derogatis & Derogatis, 1990). Kaye (1989) evaluated the impact of dialysis on the adjustment of patients from multiple treatment centers. Pre-post assessments with the PAIS revealed significant improvement on Health Care Orientation, Vocational Environment, and Extended Family Domains. DeNour, (1982) contrasted the PAIS-SR scores of dialysis patients rated as “good” and “bad” adjusters by their physicians. Statistically significant differences between groups were observed on the PAIS-SR total and PAIS-SR domain scores. Friedman (1988) and her colleagues related PAIS dimension scores to measures of family type, cohesion, and satisfaction in a sample of 57 women with breast cancer. PAIS scores significantly discriminated between family types and levels of cohesion and showed significant correlations with measures of family satisfaction and adjustment.

Convergent Validity: The following are correlations between PAIS Domains and Total Scores and Other Psychological Test Scores. Very high correlations are observed between the
PAIS total adjustment score and the score on this thermometer-type global adjustment rating scale \((r = .81)\). Although the relationship between the SCL-90-R and the PAIS total score is substantial \((r = .60)\), it is not as high as in the case of the GAIS, reflecting the fact that psychosocial adjustment is comprised of more than simply an absence of psychological symptoms. The PAIE scale was used as a simple measure of the patient’s attitudes and expectancies concerning his illness and the information he or she possessed. It correlated substantially with the PAIS Total Score \((r = .88)\).
Table 3.

*PAIS Domains and Total Scores.*

<table>
<thead>
<tr>
<th>External Criterion Measure</th>
<th>PAIS Total Score</th>
<th>Domain I (HCO)</th>
<th>Domain II (VE)</th>
<th>Domain III (DE)</th>
<th>Domain IV (SR)</th>
<th>Domain V(EFR)</th>
<th>Domain VI(SE)</th>
<th>Domain VII (PD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Global</td>
<td>.81</td>
<td>.80</td>
<td>.54</td>
<td>.76</td>
<td>.46</td>
<td>.40</td>
<td>.12</td>
<td>.70</td>
</tr>
<tr>
<td>Adjustment to Illness Scale (GAIS)</td>
<td>.60</td>
<td>.34</td>
<td>.39</td>
<td>.45</td>
<td>.13</td>
<td>.57</td>
<td>.30</td>
<td>.83</td>
</tr>
<tr>
<td>SCL-90-R</td>
<td>.69</td>
<td>.31</td>
<td>.46</td>
<td>.67</td>
<td>.42</td>
<td>.58</td>
<td>.31</td>
<td>.77</td>
</tr>
<tr>
<td>General Severity Index</td>
<td>.64</td>
<td>.88</td>
<td>.28</td>
<td>.52</td>
<td>.40</td>
<td>.06</td>
<td>.14</td>
<td>.42</td>
</tr>
<tr>
<td>Patient's Attitudes,</td>
<td>.64</td>
<td>.88</td>
<td>.28</td>
<td>.52</td>
<td>.40</td>
<td>.06</td>
<td>.14</td>
<td>.42</td>
</tr>
<tr>
<td>Information &amp; Expectancies (PAIE) Scale</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Domain I = Health Care Orientation, 
Domain II = Vocational Environment, 
Domain III = Domestic Environment, 
Domain IV = Sexual Relationships, 
Domain V = Extended Family Relationships, 
Domain VI = Social Environment, 
Domain VII = Psychological Distress.
Predictive validity refers to how well a test predicts or discriminates among outcomes, and defines the utility of the instrument for various classes of predictive applications. The PAIS and the PAIS-SR have been used in numerous studies and have confirmed their predictive capabilities relative to numerous external criteria of interest. Evans et al (1988) employed the PAIS to demonstrate clear differences in levels of adjustment between cancer patients suffering from clinical depression and treated appropriately, and similar patients who did not receive appropriate treatment. Gilbar & DeNour (1989) used the PAIS to evaluate 70 cancer patients who refused treatment with chemotherapy, and compared them with 70 matched patients who participated in the prescribed regimen. Results reflected significantly poorer adjustment on six of seven of the PAIS measures among the cohort who refused treatment, with only the Extended Family domain failing to discriminate the groups.

Data Collection Procedure

The clinic nurses identified eligible participants and asked them for permission to be contacted by the Principal Investigator (PI). Once identified, the PI obtained informed written consent during the period of August 2004 – May 2005. Participants for this study were recruited in two different settings. First, participants were recruited through the hemophilia clinic at Kapiolani Medical Center in Honolulu, Hawaii. Each participant was mailed the questionnaire packet following receipt of informed consent. Complete instructions for questionnaire completion and a return self-addressed stamped envelope were provided. Participants attending clinic during the enrollment period were given the option of completing the questionnaires at the clinic or at their convenience at home and returning them by mail. A brief explanation of the purpose of the study and directions for completing questionnaires was included in the mailing.
Enrollment of participants began after assessment of eligibility was completed and informed consent was obtained.

Secondly, recruitment of participants occurred during the annual membership meeting sponsored by the Hemophilia Foundation of Hawaii (HFH). On October 16, 2004 the HFH, a nonprofit organization whose mission is to promote the well being of persons with inherited bleeding disorders, held an annual educational symposium for its members. Participants for this study were recruited during this meeting. Informed consent was obtained during the meeting and eligible participants were mailed the questionnaire packet.

The participants were mailed the questionnaires and follow-up telephone calls were made two months later to those who had not returned the questionnaires. There were a total of 136 questionnaires that were mailed or distributed to potential participants; the response rate was 44%.

Data Analysis

The statistical package for the social sciences (SPSS) Version 12 was used for basic summary statistics (SPSS Inc., Chicago, IL, 2004). A p-value of 0.05 was used to indicate statistical significance. Data analysis consisted of descriptive statistics, correlational analysis and a series of multiple regression equations to test for mediation. Reliabilities were calculated for health related hardiness and PAIS measures in this sample. To address aim (1) describe demographics and illness variables, health stressors, health related hardiness, perception of illness impact, self perception of health status and psychosocial adjustment to illness, the data was analyzed using descriptive statistics, including the mean, standard deviations, range for continuous variables, and percentages for nominal variables. To address (aim 2) determine relationships between demographic and illness variables for categorical variables, health
stressors, health related hardiness, perception of illness impact, self-perception of health status and psychosocial adjustment to illness. A correlation matrix was developed using Pearson’s product moment correlational analysis. This examined the relationships between all study variables and the relationships of the demographic and illness variables with the main study variables. To address aim (3) determine if perception of illness impact has a direct and/or mediating effect on the relationship between health stressors, health related hardiness, and self-perception of health status, and psychosocial adjustment to illness. The test for mediation was conducted using six regression equations. First, the outcome variable PAIS or SPHS was regressed on the on the predictor variable health stressors or health related hardiness. If a significant relationship was found then we proceeded to equation two. In the second equation, the mediator variable perception of illness impact was regressed on the predictor variable health stressors or health related hardiness. The third equation involved regressing the outcome variable PAIS or self-perception of health status simultaneously on the predictor variable health stressors or health related hardiness and the mediator variable perception of illness impact.

Protection of Human Subjects

Institutional Review Board approval from the University of Hawaii Committee on Human Studies and Hawaii Pacific Health was obtained prior to implementation of the study. The study consent form and Health Insurance Portability and Accountability Act (HIPAA) forms are attached (See Appendix G).

Every subject was informed that (a) data shared with the principal investigator would be kept confidential and anonymous and (b) that research findings would be reported only as group data. The participants were assured their participation was voluntary and that if they chose not to
participate in the study that it would not affect, in any way, the care and treatment they received at the HTTCH. They could also withdraw from the study at any time without penalty.
Chapter 4: Results

Analysis

This chapter presents the analysis of data, description of the sample and findings of the study. Descriptive statistics are presented to describe the subjects’ demographics. A correlation matrix using Pearson’s product moment correlational analysis was used to determine the relationships between all the study variables and also the relationships of the demographic and illness variables with the study variables. A series of regressions was used to test for mediating the relationship of perception of illness impact.

Description of the Sample

A non-probability sample of convenience comprised the sample. Sixty participants were in the study. All participants received care from a comprehensive clinic at a federally funded hemophilia treatment center in Honolulu, Hawaii.

Demographics

Age and Gender. The mean age of the sample was 38 years (SD = 13.9) and ranged from 18 to 84 years. Male participants comprised 40% (n = 24) of the sample and female participants comprised 60% (n = 36) of the sample (See Table 4).
Table 4.

*Distribution of the Sample by Gender and Age*

<table>
<thead>
<tr>
<th>Gender</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male</td>
<td>24</td>
<td>40</td>
</tr>
<tr>
<td>Female</td>
<td>36</td>
<td>60</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Age (years) (n = 60)</th>
<th>Mean</th>
<th>SD</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>38</td>
<td>13.9</td>
<td>18-84</td>
</tr>
</tbody>
</table>
Marital Status. Half of the sample (n = 30) were single; 41.7% (n = 25) were married; 6.7% (n = 4) were divorced and, 1.7% (n = 1) were widowed (See Table 5).

Table 5.

*Distribution of the Sample by Marital Status*

<table>
<thead>
<tr>
<th>Marital Status</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Single</td>
<td>30</td>
<td>50.0</td>
</tr>
<tr>
<td>Married</td>
<td>25</td>
<td>41.7</td>
</tr>
<tr>
<td>Divorced</td>
<td>4</td>
<td>6.7</td>
</tr>
<tr>
<td>Widowed</td>
<td>1</td>
<td>1.7</td>
</tr>
</tbody>
</table>
Ethnicity. The sample represented an ethnically diverse population of Caucasian, Japanese, Asian, Filipino, Hawaiian or Part Hawaiian, and Mixed Race participants (See Table 3). One quarter of the sample was Filipino (n = 15), while Japanese (20%, n = 12) were the second highest ethnic group. Caucasians represented 18.3% of the sample (n = 11), Hawaiian or Part Hawaiian made up 16.7% (n = 10), Asian 10% (n = 6) and Mixed Race (10 %, n = 6).

Table 6.

*Distribution of the Sample by Ethnicity*

<table>
<thead>
<tr>
<th>Ethnicity</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Filipino</td>
<td>15</td>
<td>25.0</td>
</tr>
<tr>
<td>Japanese</td>
<td>12</td>
<td>20.0</td>
</tr>
<tr>
<td>Caucasian</td>
<td>11</td>
<td>18.3</td>
</tr>
<tr>
<td>Hawaiian/Part Hawaiian</td>
<td>10</td>
<td>16.7</td>
</tr>
<tr>
<td>Asian*</td>
<td>6</td>
<td>10.0</td>
</tr>
<tr>
<td>Mixed Race</td>
<td>6</td>
<td>10.0</td>
</tr>
</tbody>
</table>

* Indicates Chinese, Korean, Vietnamese/Other Southeast Asian
Education. Only 6.6% (n = 4), had not completed high school with almost half 46.7% (n = 28), of the sample reporting completion of a high school education. Sample participants who had completed college were 38% (n = 23); 6.7% (n = 4) had completed graduate school and, one participant had completed a doctorate 1.7% (n = 1) (See Table 7).

Table 7.

**Distribution of the Sample by Education**

<table>
<thead>
<tr>
<th>Educational Level</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Elementary School</td>
<td>2</td>
<td>3.3</td>
</tr>
<tr>
<td>Junior High School</td>
<td>2</td>
<td>3.3</td>
</tr>
<tr>
<td>High School</td>
<td>28</td>
<td>46.7</td>
</tr>
<tr>
<td>College</td>
<td>23</td>
<td>38.3</td>
</tr>
<tr>
<td>Graduate School</td>
<td>4</td>
<td>6.7</td>
</tr>
<tr>
<td>Doctorate</td>
<td>1</td>
<td>1.7</td>
</tr>
</tbody>
</table>
**Employment status.** More participants were employed than were unemployed, 70% (n = 42) and 30% (n = 18) respectively. More than half (76%) of the employed participants were working full-time (n = 32), and 24% (n = 10), were working part-time. Participants (n = 18; 30%) who reported no employment included homemakers and students (See Table 8).

**Table 8.**

*Distribution of the Sample by Employment and Employment Time (ET)*

<table>
<thead>
<tr>
<th>Employment and ET</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Yes</td>
<td>42</td>
<td>70.0</td>
</tr>
<tr>
<td>Full-time</td>
<td>32</td>
<td>76.0</td>
</tr>
<tr>
<td>Part-time</td>
<td>10</td>
<td>24.0</td>
</tr>
<tr>
<td>No</td>
<td>18</td>
<td>30.0</td>
</tr>
</tbody>
</table>
Annual Income. Participants’ annual income ranged from less than $10,000 to more than $65,000. The median income was slightly over $20,000. The majority of the sample (n = 45; 75%) had annual incomes of less than $35,000 (See Table 9).

Table 9.

Distribution of the Sample by Income

<table>
<thead>
<tr>
<th>Annual Income</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>$10,000 or less</td>
<td>17</td>
<td>28.3</td>
</tr>
<tr>
<td>$10,001 - $20,000</td>
<td>12</td>
<td>20.0</td>
</tr>
<tr>
<td>$20,001 - $35,000</td>
<td>16</td>
<td>26.7</td>
</tr>
<tr>
<td>$35,001 - $50,000</td>
<td>7</td>
<td>11.7</td>
</tr>
<tr>
<td>$50,001 - $65,000</td>
<td>5</td>
<td>8.3</td>
</tr>
<tr>
<td>Over 65,001</td>
<td>3</td>
<td>5.0</td>
</tr>
</tbody>
</table>
Number of Children. Forty percent (n = 24) did not have children. Slightly more than half 53.4% of the participants had 1 – 3 children (n = 32) and 6.6% (n = 4) had 4 - 6 children (See Table 10).

Table 10.

Distribution of the Sample by Number of Children

<table>
<thead>
<tr>
<th>Children</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>24</td>
<td>40.0</td>
</tr>
<tr>
<td>1-3</td>
<td>32</td>
<td>53.4</td>
</tr>
<tr>
<td>4-6</td>
<td>4</td>
<td>6.6</td>
</tr>
</tbody>
</table>
*Relatives in the home.* The number of relatives in the home of participants ranged from 0 – 11 with slightly over half of the sample (n = 31; 51.7%) having no relatives living with them. There were 22 sample participants who had 1 – 3 relatives living with them. The mean number of relatives living in the house was 1.38 (SD = 2.03, range 0 – 11) (See Table 11).

Table 11.

*Distribution of Others in the Home (Relatives)*

<table>
<thead>
<tr>
<th># of Relatives</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>31</td>
<td>51.7</td>
</tr>
<tr>
<td>1</td>
<td>8</td>
<td>13.3</td>
</tr>
<tr>
<td>2</td>
<td>6</td>
<td>10.0</td>
</tr>
<tr>
<td>3</td>
<td>8</td>
<td>13.3</td>
</tr>
<tr>
<td>4</td>
<td>3</td>
<td>5.0</td>
</tr>
<tr>
<td>5</td>
<td>2</td>
<td>3.3</td>
</tr>
<tr>
<td>6</td>
<td>1</td>
<td>1.7</td>
</tr>
<tr>
<td>11</td>
<td>1</td>
<td>1.7</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th># of Relatives (n = 60)</th>
<th>Mean</th>
<th>SD</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.38</td>
<td>2.03</td>
<td>0-11</td>
<td></td>
</tr>
</tbody>
</table>
Non-Relatives in the Home. The number of non-relatives living in the home ranged from 0 - 4 with over three-fourths (n = 48; 80.0%) having no non-relatives in the home. There were 11 sample participants (18.3%) who had 1 – 2 non-relatives living in the home. The mean number of non-relatives living in the house was .30 (SD = .72, Range 0 – 4) (See Table 12).

Table 12.

Distribution of Others Living in the Home (Non-Relatives)

<table>
<thead>
<tr>
<th># of Non-Relatives</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>48</td>
<td>80.0</td>
</tr>
<tr>
<td>1</td>
<td>8</td>
<td>13.3</td>
</tr>
<tr>
<td>2</td>
<td>3</td>
<td>5.0</td>
</tr>
<tr>
<td>4</td>
<td>1</td>
<td>1.7</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th># of Non-Relatives (n = 60)</th>
<th>Mean</th>
<th>SD</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>.30</td>
<td>.72</td>
<td>0 - 4</td>
</tr>
</tbody>
</table>
Type of Bleeding Disorders. Forty percent (n = 24) were hemophilia carriers, 36.7% (n = 22) had hemophilia, 20% (n = 12) had von Willebrands disease and, 3.3% (n = 2) had factor V deficiency (See Table 13).

Table 13.

*Distribution of Sample by Bleeding Disorder Type and Gender*

<table>
<thead>
<tr>
<th>Bleeding Disorder</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hemophilia Carriers</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Men</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Women</td>
<td>24</td>
<td>40.0</td>
</tr>
<tr>
<td>Hemophilia</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Men</td>
<td>22</td>
<td>36.7</td>
</tr>
<tr>
<td>Women</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>von Willebrands Disease</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Men</td>
<td>2</td>
<td>3.3</td>
</tr>
<tr>
<td>Women</td>
<td>10</td>
<td>16.7</td>
</tr>
<tr>
<td>Factor V</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Men</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Women</td>
<td>2</td>
<td>3.3</td>
</tr>
</tbody>
</table>
Additional Health Conditions/Chronic Illnesses. There were 13 other health conditions in addition to the bleeding disorders that were present in the sample. A quarter of the participants (n = 15) had hepatitis C (25%). The mean number of chronic illnesses was 2.18 (SD = 1.4) with a range of 1 – 6 (See Table 14). Twenty-two or 36.7% of participants had only the bleeding disorder and no other chronic illness were reported. Thirty-eight or 63.3% reported more than one chronic illness.
Table 14.

*Distribution of Sample by Other Health Conditions*

<table>
<thead>
<tr>
<th>Other Health Conditions</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hepatitis C</td>
<td>15</td>
<td>25</td>
</tr>
<tr>
<td>High Blood Pressure</td>
<td>10</td>
<td>16.7</td>
</tr>
<tr>
<td>Asthma</td>
<td>9</td>
<td>15</td>
</tr>
<tr>
<td>Arthritis</td>
<td>7</td>
<td>11.7</td>
</tr>
<tr>
<td>Heart Disease</td>
<td>6</td>
<td>10</td>
</tr>
<tr>
<td>Hepatitis B</td>
<td>5</td>
<td>8.3</td>
</tr>
<tr>
<td>HIV/AIDS</td>
<td>5</td>
<td>8.3</td>
</tr>
<tr>
<td>Diabetes</td>
<td>4</td>
<td>6.7</td>
</tr>
<tr>
<td>Hepatitis A</td>
<td>3</td>
<td>5.0</td>
</tr>
<tr>
<td>Cancer</td>
<td>2</td>
<td>3.3</td>
</tr>
<tr>
<td>Hypothyroidism</td>
<td>1</td>
<td>1.7</td>
</tr>
<tr>
<td>Iron Deficiency Anemia</td>
<td>1</td>
<td>1.7</td>
</tr>
<tr>
<td>Kidney Disease</td>
<td>1</td>
<td>1.7</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Number of Chronic Illnesses (n = 60)</th>
<th>Mean</th>
<th>SD</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>2.18</td>
<td>1.4</td>
<td>1 - 6</td>
</tr>
</tbody>
</table>
**Aim 1:** To describe health stressors, health related hardiness, perception of illness impact, self-perception of health status and psychosocial adjustment to illness in individuals diagnosed with inherited bleeding disorders.

**Descriptive Statistics for Main Study Variables**

The sample mean for health stressors was 2.18 (SD = 1.4, Range 1 - 6). The sample mean for health related hardiness was 148.77 (SD = 14.89, Range 120 - 180). The sample mean for perception of illness impact was 3.98 (SD = 3.39, Range 1 - 10). The sample mean for self-perception of health was 2.77 (SD = 0.72, Range 1 - 4). The sample mean for the PAIS-SR total score was 350 (SD = 50.15, Range 263-486). The sample mean for the PAIS-SR total T-score was 50.38 (SD = 10.51, Range 30 - 80) (See Table 15). Ten participants had a total T-score (n = 10 or 17%) of 62 or greater. “As a rough rule to determine “caseness”, preliminary analyses have suggested that respondents with a PAIS total score equivalent to or greater than a T-score of 62 are positive for clinical levels of psychosocial maladjustment” (Derogatis & Derogatis, 1990). Reliability (Cronbach’s alpha) for the HRHS Total Score, .74, for the PAIS-SR Total raw score, .91 and for the PAIS total T-score, .79 (See Table 15).
Table 15.

*Descriptive Statistics for Main Study Variables*

<table>
<thead>
<tr>
<th>Study Variables</th>
<th>Mean</th>
<th>SD</th>
<th>Sample Range</th>
<th>Possible Range</th>
<th>Cronbach’s Alpha</th>
</tr>
</thead>
<tbody>
<tr>
<td>Health Stressors</td>
<td>2.18</td>
<td>1.40</td>
<td>1 - 6</td>
<td>1 - ~</td>
<td>N/A</td>
</tr>
<tr>
<td>HRHS (Total Score)</td>
<td>148.77</td>
<td>14.89</td>
<td>120 - 180</td>
<td>34 - 204</td>
<td>.74</td>
</tr>
<tr>
<td>Perception of Illness Impact</td>
<td>3.98</td>
<td>3.39</td>
<td>1 - 10</td>
<td>1 - 10</td>
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<tr>
<td>Self-Perception of Health</td>
<td>2.77</td>
<td>.72</td>
<td>1 - 4</td>
<td>1 - 4</td>
<td>N/A</td>
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<tr>
<td>PAIS Total Raw Score</td>
<td>350</td>
<td>50.15</td>
<td>263 - 486</td>
<td>263 - 560</td>
<td>.91</td>
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<tr>
<td>PAIS Total T-Score**</td>
<td>50.38</td>
<td>10.51</td>
<td>30-80</td>
<td>30 - 80*</td>
<td>.79</td>
</tr>
</tbody>
</table>

~ Infinite

* Range for Normative Diabetic Group

**When evaluating groups as opposed to individuals, if there is no interest in comparing a patient sample of one of the published normative groups, raw scores will function adequately in any inferential statistical analyses. In fact, if normative comparisons are not of interest, raw scores are actually a bit more precise since they avoid the rounding error associated with standardized scores.* (Derogatis & Derogatis, 1990). No normative group was available for individuals with inherited bleeding disorders.
Table 16 presents sample mean scores for the PAIS total raw-scores and the mean scores for the PAIS total T-scores in each of the seven domains. These seven domains are: 1) Health care orientation 2) Vocational environment 3) Domestic environment 4) Sexual relationships 5) Extended family relationships 6) Social environment and 7) Psychological distress. The sample mean and standard deviations for both the PAIS raw score and T-Score on each domain on the PAIS-SR are presented in Table 16.
Table 16.

*Descriptive Statistics for PAIS-SR*

N = 60

<table>
<thead>
<tr>
<th>Domains</th>
<th>Sample Raw Score</th>
<th>Sample T-Score</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean (SD)</td>
<td>Mean (SD)</td>
</tr>
<tr>
<td>Health Care Orientation</td>
<td>5.77 (3.35)</td>
<td>55.93 (13.18)</td>
</tr>
<tr>
<td>(8 items)</td>
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<td></td>
</tr>
<tr>
<td>Vocational Environment</td>
<td>3.29 (4.01)</td>
<td>50.24 (11.16)</td>
</tr>
<tr>
<td>(6 items) (n = 59)*</td>
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<td></td>
</tr>
<tr>
<td>Domestic Environment</td>
<td>4.00 (4.35)</td>
<td>47.53 (11.70)</td>
</tr>
<tr>
<td>(8 items)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sexual Relationships</td>
<td>2.63 (4.33)</td>
<td>48.58 (9.24)</td>
</tr>
<tr>
<td>(6 items) (n = 57)*</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Extended Family Relations</td>
<td>1.13 (2.93)</td>
<td>50.00 (8.41)</td>
</tr>
<tr>
<td>(5 items)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Social Environment</td>
<td>3.28 (4.46)</td>
<td>50.78 (9.55)</td>
</tr>
<tr>
<td>(6 items)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Psychological Distress</td>
<td>4.82 (4.14)</td>
<td>46.72 (9.83)</td>
</tr>
<tr>
<td>(7 items)</td>
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<td></td>
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</tbody>
</table>

* n varied due to missing data
Aim 2: Determine relationships between demographic and illness variables, health stressors, health related hardiness, perception of illness impact, self-perception of health status and psychosocial adjustment to illness.

A correlation matrix was completed between the demographic and illness variables (See Table 14). There was a positive correlation ($r = .49, p < .01$) between age and number of children; a negative correlation ($r = -.26, p < .05$) between age and the number of relatives in the home; and a positive correlation ($r = .40, p < .01$) between age and years since diagnosis. Older participants had more children, fewer relatives living in the home and had been diagnosed longer with their bleeding disorder. There was a positive correlation ($r = .26, p < .05$) between gender (females) and the number of children; and a positive correlation ($r = .64, p < .01$) between gender and bleeding disorder. There was a negative correlation ($r = -.48, p < .01$) between gender (males) and years since diagnosis. Females had more children; were more likely to be diagnosed as carriers and had been diagnosed a shorter time than males. There was a negative correlation ($r = -.41, p < .01$) between marital status and education. Married participants had higher education. There was a negative correlation ($r = -.27, p < .05$) between employment-time and income. There was a positive correlation ($r = .31, p < .05$) between income and number of children. Higher income participants had more children. There is a negative correlation ($r = -.40, p < .01$) between bleeding disorder and years since diagnosis. Participants with hemophilia (males) had been diagnosed longer.

A correlation matrix depicts the relationship between demographic, and main study variables (See Table 17). Age was positively correlated with health stressors ($r = .34, p < .01$); and years since diagnosis ($r = .46; p < .01$). Gender (males) was negatively correlated with health stressors ($r = -.33; p < .01$) (males had higher health stressors); and with perception of illness
impact ($r = -0.63, p <.01$), positively correlated with self-perception of health status ($r = 0.26, p < .05$) and negatively correlated with psychosocial adjustment to illness ($r = -0.41, p < .01$). There was a negative correlation ($r = -0.30, p <.05$) between number of children and psychosocial adjustment to illness. There was a negative correlation ($r = -0.34, p <.01$) between type of bleeding disorder and perception of illness impact. Years since diagnosis was positively correlated with ($r = -0.46, p <.01$) health stressors; negatively correlated with ($r = -0.31, p < .05$) health related hardiness; positively correlated with ($r = 0.57, p <.01$) perception of illness impact and positively correlated with ($r = 0.62, p <.01$) psychosocial adjustment to illness.

In summary, older male participants who had been diagnosed longer with their bleeding disorder reported more health stressors. Males with hemophilia who had been diagnosed longer also reported higher perception of illness impact. Males overall had poorer psychosocial adjustment to illness as did those who had been diagnosed longer with their bleeding disorder. Participants who had been diagnosed longer also had lower health related hardiness. However, having children was associated with better psychosocial adjustment to illness.

A correlation matrix depicts the relationships between the main study variables (See Table 19). Health stressors were positively correlated with perception of illness impact ($r = 0.35; p < .01$); negatively correlated with self-perception of health status ($r = -0.58; p < .01$) and positively correlated with psychosocial adjustment to illness (raw scores; $r = 0.50; p < .01$). Health related hardiness was negatively correlated with perception of illness impact ($r = -0.30; p < .05$); positively correlated with self-perception of health status ($r = 0.36; p = <.01$); and negatively correlated with psychosocial adjustment to illness (raw score, $r = -0.29; p < .05$). Perception of illness impact was positively correlated with psychosocial adjustment to illness (raw score; $r =$
Self-perception of health status and psychosocial adjustment to illness were negatively correlated (r = -.45; p < .01).

In summary, higher health stressors were associated with higher perception of illness impact, lower perception of health status and poorer psychosocial adjustment to illness. Higher health related hardiness was associated with a lower perception of illness impact, higher self-perception of health status, and better psychosocial adjustment to illness. Higher perception of illness impact was associated with lower self-perception of health status and poorer psychosocial adjustment to illness. Higher self-perception of health status was associated with better psychosocial adjustment to illness.
Table 17.

*Correlations Between Demographic Variables*

<table>
<thead>
<tr>
<th></th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>11</th>
<th>12</th>
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</thead>
<tbody>
<tr>
<td>1. Age</td>
<td>X</td>
<td>-0.12</td>
<td>-0.16</td>
<td>0.08</td>
<td>0.04</td>
<td>0.17</td>
<td>0.49**</td>
<td>-0.26*</td>
<td>-0.08</td>
<td>-0.12</td>
<td>0.40**</td>
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<td>2. Gender</td>
<td>X</td>
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<td>-0.19</td>
<td>-0.06</td>
<td>-0.07</td>
<td>-0.20</td>
<td>0.26*</td>
<td>-0.05</td>
<td>0.06</td>
<td>0.64**</td>
<td>-0.48**</td>
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</tr>
<tr>
<td>3. Marital Status</td>
<td>X</td>
<td>-0.41**</td>
<td>-0.07</td>
<td>-0.05</td>
<td>0.06</td>
<td>-0.15</td>
<td>0.18</td>
<td>0.13</td>
<td>0.12</td>
<td>-0.05</td>
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<tr>
<td>4. Education</td>
<td>X</td>
<td>-0.10</td>
<td>-0.12</td>
<td>-0.03</td>
<td>-0.06</td>
<td>-0.08</td>
<td>0.12</td>
<td>-0.19</td>
<td>0.02</td>
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<tr>
<td>5. Employment</td>
<td>X</td>
<td>0.88**</td>
<td>-0.23</td>
<td>-0.03</td>
<td>0.15</td>
<td>-0.17</td>
<td>-0.01</td>
<td>-0.05</td>
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</tr>
<tr>
<td>6. Employment-time</td>
<td>X</td>
<td>-0.27*</td>
<td>-0.13</td>
<td>0.11</td>
<td>-0.19</td>
<td>-0.01</td>
<td>-0.02</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>7. Income</td>
<td>X</td>
<td>0.31*</td>
<td>-0.18</td>
<td>0.02</td>
<td>0.13</td>
<td>-0.08</td>
<td></td>
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<td></td>
</tr>
<tr>
<td>8. # of Children</td>
<td>X</td>
<td>-0.10</td>
<td>-0.18</td>
<td>0.19</td>
<td>-0.23</td>
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<td></td>
</tr>
<tr>
<td>9. # of Home Relatives</td>
<td>X</td>
<td>0.13</td>
<td>0.07</td>
<td>-0.17</td>
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<td></td>
<td></td>
<td></td>
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<tr>
<td>10. # of Non-relatives</td>
<td>X</td>
<td>0.25</td>
<td>-0.04</td>
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<tr>
<td>11. Bleeding Disorder</td>
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<td>-0.40**</td>
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<tr>
<td>12. Years since Diagnosis</td>
<td>X</td>
<td></td>
<td></td>
<td></td>
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<td></td>
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</tbody>
</table>

* * p = <.05  
** ** p = <.01  

*Correlations Between Demographic Variables*
Table 18.

Correlations Between Demographic and Main Study Variables (* \( p < .05 \), ** \( p < .01 \))

<table>
<thead>
<tr>
<th></th>
<th>Health Stressors</th>
<th>HRHS</th>
<th>PIIS</th>
<th>SPHS</th>
<th>PAIS Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Age</td>
<td>.34**</td>
<td>-.01</td>
<td>-.03</td>
<td>-.25</td>
<td>-.60</td>
</tr>
<tr>
<td>2. Gender</td>
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<td>-.20</td>
<td>-.63**</td>
<td>.26*</td>
<td>-.41**</td>
</tr>
<tr>
<td>3. Marital Status</td>
<td>-.01</td>
<td>.04</td>
<td>-.15</td>
<td>.05</td>
<td>-.12</td>
</tr>
<tr>
<td>4. Employment</td>
<td>.15</td>
<td>-.10</td>
<td>.05</td>
<td>.01</td>
<td>.04</td>
</tr>
<tr>
<td>5. Employment -Time</td>
<td>.15</td>
<td>-.09</td>
<td>.12</td>
<td>.05</td>
<td>.02</td>
</tr>
<tr>
<td>6. Income</td>
<td>.12</td>
<td>.11</td>
<td>.06</td>
<td>-.07</td>
<td>-.05</td>
</tr>
<tr>
<td>7. # Of Children</td>
<td>-.13</td>
<td>.16</td>
<td>-.36</td>
<td>.02</td>
<td>-.30*</td>
</tr>
<tr>
<td>8. # of Home Relatives</td>
<td>-.22</td>
<td>-.02</td>
<td>.16</td>
<td>.21</td>
<td>.08</td>
</tr>
<tr>
<td>9. # of Non-Relatives</td>
<td>-.01</td>
<td>-.07</td>
<td>.01</td>
<td>-.16</td>
<td>.09</td>
</tr>
<tr>
<td>10. Bleeding Disorder</td>
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<td>.07</td>
<td>-.34**</td>
<td>.13</td>
<td>-.14</td>
</tr>
<tr>
<td>11. Years Since Diagnosis</td>
<td>.46**</td>
<td>-.31*</td>
<td>.57**</td>
<td>-.26</td>
<td>.62**</td>
</tr>
</tbody>
</table>
Table 19.

Correlations Between Main Study Variables

<table>
<thead>
<tr>
<th></th>
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<th>16</th>
<th>17</th>
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<tbody>
<tr>
<td>13. Health Stressors</td>
<td>X</td>
<td>-.15</td>
<td>.35**</td>
<td>-.58**</td>
<td>.50**</td>
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<td>14. HRHS</td>
<td>X</td>
<td>-.30*</td>
<td>.36**</td>
<td>-.29*</td>
<td></td>
</tr>
<tr>
<td>15. PIIS</td>
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<td>-.36**</td>
<td>.66**</td>
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<td>16. SPHS</td>
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</table>

HRHS = Health Related Hardiness

PIIS = Perception of Illness Impact

SPHS = Self-perception of Health Status

PAIS -SR = Psychosocial Adjustment Illness Scale Self-Report

* p = <.05

** p = <.01
Aim 3: To determine if perception of illness impact has a direct and/or mediating effect on the relationship between health stressors, health related hardiness, and self-perception of health status, and psychosocial adjustment to illness.

First hypotheses 1 through 4 were tested with a simple linear regression analysis regarding the relationship between the independent variables (health stressors and health related hardiness) and the dependent variables (psychosocial adjustment to illness and the self-perception of health status). Secondly, hypotheses 5 and 6 were tested regarding the relationship between the mediating variable (perception of illness impact) and, the dependent variables (psychosocial adjustment to illness and self-perception of health status). Lastly, hypotheses 7, 8, 9 and 10 were tested to determine if perception of illness impact mediated the relationship between the independent variables (health stressors, health related hardiness) and the dependent variables (psychosocial adjustment to illness and self-perception of health status).

Hypothesis 1. Health stressors will be positively associated with psychosocial adjustment to illness. This hypotheses was supported; health stressors were positively associated with psychosocial adjustment to illness (b = 19.49, p = <.0000) (See Table 20 Model 1).

Hypothesis 2. Health stressors will be negatively associated with self-perception of health status. This hypothesis was supported; health stressors were negatively associated with self-perception of health status (b = -0.299, p = <.0000) (See Table 20 Model 3).

Hypothesis 3. Health related hardiness will be negatively associated with psychosocial adjustment to illness. This hypotheses was supported; health related hardiness was negatively associated with psychosocial adjustment to illness (b = -0.74, p = <.02) (See Table 20 Model 2).
**Hypothesis 4.** Health related hardiness will be positively associated with self-perception of health status. This hypothesis was supported; health related hardiness was positively associated with self-perception of health status (b = 0.013, p = < .005) (See Table 20 Model 4).

**Hypothesis 5.** Perception of illness impact will be positively associated with psychosocial adjustment to illness. This hypothesis was supported; perception of illness impact was positively associated with psychosocial adjustment to illness (b = 9.68, p = < 0.0000) (See Table 20 Models 1 and 2).

**Hypothesis 6.** Perception of illness impact will be negatively associated with self-perception of health status. This hypothesis was supported; perception of illness impact was negatively associated with self-perception of health status (b = -0.075, p = < .01) (See Table 20 Models 3 and 4).

**Hypothesis 7.** Perception of illness impact will mediate the relationship between health stressors and psychosocial adjustment to illness. This hypothesis was not supported (See Table 20 Model 1). When perception of illness impact is the mediator in the relationship between health stressors and psychosocial adjustment to illness (b = 7.83; p = < .0000) the relationship between health stressors and psychosocial adjustment to illness remains significant (b = 12.80; p = < .005).

**Hypothesis 8.** Perception of illness impact will mediate the relationship between health related hardiness and psychosocial adjustment to illness. This hypothesis was supported. (See Table 20 Model 2). When perception of illness impact is the mediator in the relationship between health related hardiness and psychosocial adjustment to illness (b = 9.17; p = < .0000), the relationship between health related hardiness and psychosocial adjustment to illness then became non-significant (b = -.29; p = < .26) (See Table 20 Model 2).
Hypothesis 9. Perception of illness impact will mediate the relationship between health stressors and self-perception of health. This hypothesis was not supported (See Table 20 Model 3). When the perception of illness impact is the mediator in the relationship between health stressors and self-perception of health status (b = -0.037; p = <.13); the mediating relationship is non-significant and the relationship between health stressors and the self-perception of health status remains significant (b = -0.268 ; p = <.0000).

Hypothesis 10. Perception of illness impact will mediate the relationship between health related hardiness and self-perception of health. This hypothesis was not supported (See Table 20 Model 4). When the perception of illness impact is the mediator in the relationship between health related hardiness and self-perception of health (b = -0.058; p = < .03), the relationship between health stressors and self-perception of health status remains significant (b = -0.010; p = < .03).

In summary, for models 1, 3, and 4 the effect of the independent variable (health stressors and health related hardiness respectively) on the dependent variables (psychosocial adjustment to illness and self-perception of health status) did not change with the mediator. However, in model 2 the effect of the independent variable (health related hardiness) on the dependent variable (psychosocial adjustment to illness) did change with the mediator; with perception of illness impact as the mediator, the relationship between health related hardiness and psychosocial adjustment to illness became nonsignificant (p = <.26).
Table 20.

*Results of Mediation Models*

<table>
<thead>
<tr>
<th>Model #</th>
<th>IV</th>
<th>Mediator</th>
<th>DV</th>
<th>b</th>
<th>p</th>
<th>beta</th>
<th>p Conclusion</th>
</tr>
</thead>
<tbody>
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<td>PAIS Total</td>
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<tr>
<td></td>
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<td>PIIS</td>
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<tr>
<td></td>
<td>PIIS</td>
<td>PAIS Total</td>
<td>9.68</td>
<td>.0000</td>
<td>0.66</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>HS</td>
<td>PAIS Total</td>
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<td>.0005</td>
<td>0.36</td>
<td>.0000</td>
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<td></td>
<td>PIIS</td>
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<tr>
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<tr>
<td></td>
<td>HRH</td>
<td>PAIS Total</td>
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<td>-0.12</td>
<td>.0000</td>
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<tr>
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<td>0.62</td>
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*IV = Independent Variable  DV = Dependent Variable*
### Results of Mediation Models

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<th>Model #</th>
<th>IV</th>
<th>Mediator</th>
<th>DV</th>
<th>b</th>
<th>p</th>
<th>beta</th>
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<tr>
<td></td>
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*HS* = Health Stressors; *PIIS* = Perception of Illness Impact; *PAIS* = Psychosocial Adjustment to Illness; *HRH* = Health Related Hardiness and *SPHS* = Self-Perception of Health Status
Chapter 5: Discussion

The discussion of this study will be guided by the aims and hypotheses of the study, the study model drawn from Pollock's research, and prior research from the literature. Limitations as well as strengths of the study will be addressed followed by implications for nursing practice and recommendations for future research. The following is a discussion of the descriptive statistics and it will address significant as well as non-significant findings.

Aim 1: To describe health stressors, health related hardiness, perception of illness impact, self-perception of health status and psychosocial adjustment to illness in individuals diagnosed with inherited bleeding disorders.

Demographics and Study Variables

The sample had a greater number of female participants than males. The genetics of hemophilia make clear that female participants carry the defective gene and their male and female offspring have a fifty-fifty chance of inheriting the disease. Males would have been diagnosed earlier because clinically many who are moderate or severe hemophiliacs are diagnosed at birth during circumcision. A small percentage of mild hemophiliacs may be diagnosed later in life if there is no known family history and they experience no bleeding problems. Some adults who are diagnosed later in life often present with bleeding problems secondary to surgical intervention, trauma or tooth extractions. Females whose fathers have hemophilia will be obligate carriers of hemophilia meaning all females born to men with hemophilia will automatically be hemophilia carriers. Females in this sample had more children and had been diagnosed a shorter time than males. Females may be diagnosed later in life unless they have bleeding problems or are symptomatic carriers they may not be tested for carrier status. Females may not undergo genetic testing for various reasons: cost (insurance companies
do not cover genetic testing); no interest in obtaining information; many females may postpone being tested until they decide to get married or have children. One of the disadvantages of getting tested earlier in life is being diagnosed with a chronic illness may jeopardize or increase health insurance premiums. The genetics of von Willebrand Disease show that this disease affects males and females equally. Diagnoses may be delayed unless individuals are symptomatic. The cost of specialized blood testing that is required for accurately diagnosing vWD is high. In the state of Hawaii no laboratory has the capacity to perform some of the specialized testing and specimens need to be sent to the mainland. Some of the tests are often repeated several times before an accurate diagnosis can be made. Males with vWD may be worked up for hemophilia because the signs and symptoms are similar.

The oldest participant was 84 years of age showing that persons with bleeding disorders can have a long life span. It makes sense that symptomatic individuals with bleeding disorders are living longer due to the introduction of home therapy and prophylaxis. Almost all of the sample had completed high school with almost one-half having a college education. This indicates that sample participants with inherited bleeding disorders were able to complete their education. A larger number of sample participants were employed. More than half of the sample was employed full-time. This may be related to economic necessity in Hawaii. Hawaii’s hemophilia treatment center continues to have the highest proportion of employed adults in the region (California, Nevada & Guam).

Almost 82% of the sample was of Asian Pacific Islander ethnicity. This is the first psychosocial study on inherited bleeding disorders that has utilized a predominantly Asian/Pacific Islander ethnic population. The majority of the sample made less than $35,000 per year. Regardless of income, individuals in the state of Hawaii can obtain health insurance
through one of the state funded programs. The state of Hawaii requires that employers provide health insurance for employees working at least twenty hours per week. Therefore, almost all individuals have some form of health insurance.

Some participants in the sample were managing one or more chronic illnesses and several had a maximum of six chronic illnesses they were managing simultaneously. Results indicate that participants in this sample had a moderate to high level of hardiness. Mean score for the perception of illness impact reflected a moderate impact but the sample range reflected minimum to maximum (a great deal). On average participants rated their health fair to good.

PAIS raw scores in the study sample were similar in terms of mean and range to the PAIS raw scores from the diabetic comparison group. The majority of the sample was not in the range for clinical psychosocial maladjustment; however, 10 individuals were at risk for psychosocial maladjustment. The PAIS may be a valuable tool for evaluating persons with inherited bleeding disorders and potentially identifying those that may be at risk for psychosocial maladjustment.

Aim 2: To determine the relationships between demographic illness variables, health stressors, health related hardiness, perception of illness impact, self-perception of health status and psychosocial adjustment to illness.

Research Question: What is the relationship between demographic and illness variables, health stressors, health related hardiness perception of illness impact, self-perception of health status and psychosocial adjustment to illness?

Hypothesis 1. Health stressors will be negatively associated with psychosocial adjustment to illness; this hypothesis was supported. The more chronic illnesses an individual had in this study, the poorer his or her psychosocial adjustment. Dealing with more than one chronic illness simultaneously can add tremendous stress in an individual’s life. This may be
attributed to increased hospitalizations and more frequent interactions with health care providers. Dealing with multiple health care providers can increase an individual’s perception of illness impact. Measurement of their psychosocial adjustment, which encompassed health care orientation (interacting frequently with the health care system), vocational environment (potential missed days at work), domestic environment (financial concerns secondary to cost of medications), sexual relationship (fear of unknown viruses), extended family relationships (financial worries related to cost of illness and care), social environment (interacting with home-care companies and managing drug shortages), and psychological distress (stigma associated with disease). This is consistent with the study findings (Pollock, Christian & Sands, 1990), which suggested that although each chronic illness has disease specific physiologic changes, the nature of the psychological adaptation process is similar. Correlations between the main study variables were all in the expected direction.

Hypothesis 2. Health stressors will be negatively associated with self-perception of health status; this hypothesis was supported. Those individuals who had higher health stressors had a lower self-perception of health status. In other words, the more chronic illnesses an individual had, the poorer his or her perception of health status. Higher health stressors could be accompanied by more physical symptoms, taking medications that may have unpleasant side effects, and reduced energy to carry out activities of daily living and independent activities of daily living. There is also the possibility of increased visits to health care providers.

Hypothesis 3. Health related hardiness will be negatively associated with psychosocial adjustment to illness; this hypothesis was supported. Those individuals who had higher health related hardiness had better psychosocial adjustment to their illness. Individuals with higher health related hardiness would have more confidence and sense of mastery over their chronic
illness (control). These individuals may then view the health stressors as an opportunity for growth (challenge), which can result in motivation and ability to deal with the illnesses more effectively (commitment). These results are consistent with Pollock’s work (Pollock, 1986) supporting the relationships between health related hardiness and psychosocial adjustment to illness. Pollock (1989a) reported that in samples of individuals with various chronic illnesses (diabetes, hypertension, and arthritis), significant relationships were found between the health related hardiness and the various psychosocial domains. In this prior study, hardiness positively influenced role function in the diabetes and hypertensive groups, social support in the arthritic and hypertensive groups, and intrapsychic functioning in the arthritic and diabetic group. These findings suggest that specific psychosocial activities may be facilitated by the presence of hardiness (Pollock, 1989a). Pollack (1989b) reported a study with 244 adults who self described as being healthy (yet many had chronic illness) where health related hardiness was positively associated with better perceived health status, use of social support and engagement in health promotion activities however, no reference was given for this study.

Hypothesis 4. Health related hardiness will be positively associated with self-perception of health status. The result of hypothesis four supports the relationship that those participants who had higher health related hardiness also had a higher self-perception of health status. Nicholas (1993, 1999) also reported a positive relationship between hardiness and self-perception of health status in older adults.

Hypothesis 5. Perception of illness impact will be positively associated with psychosocial adjustment to illness. The higher the scores on perception of illness impact, the longer the participants had been diagnosed. In this sample, those individuals who felt more impact of their illness had poorer psychosocial adjustment to their illness. Perception of illness
impact due to inherited bleeding disorders and other chronic illnesses could include: missed days at work, increased financial burden secondary to cost of treatment, more frequent disruption of family relationships, increased stress and burden in social situations and more psychological distress. This could also mean less energy, more pain, and less physical mobility.

**Hypothesis 6.** Perception of illness impact will be negatively associated with self-perception of health status; this hypothesis was supported. Participants who scored higher on the perception of illness impact were more likely to have a lower self-perception of health status. Pollock (1993) also noted that the perception of the illness impact was a critical factor and a better index of adjustment than the actual type or severity of the chronic illnesses. In summary, those participants who thought their health was good or excellent also reported that they were less affected by their illness.

**Aim 3:** To determine if perception of illness impact has a direct and/or mediating effect on the relationship between health stressors, health related hardiness, and self-perception of health status, and psychosocial adjustment to illness. The hypotheses that were not supported were 7, 9 and 10.

**Research Question:** Does perception of illness impact the relationship between health stressors and psychosocial adjustment to illness?

**Hypothesis 7.** Perception of illness impact will mediate the relationship between health stressors and psychosocial adjustment to illness; this hypothesis was not supported. The results showed no mediation of the perception of illness impact variable on the relationship between health stressors and psychosocial adjustment to illness. Neither hypothesis 7 or 9 were supported this may be due to the way in which health stressors were measured. Both the measurement of health stressors and the distribution of health stressors in the sample may have contributed to this
hypothesis not being supported. In addition, slightly over two-thirds of the sample (36.7%) only had the bleeding disorder and no other chronic illness. These individuals would most likely perceive lower impact of the illness on their daily lives than those with more than one chronic illness. The direct relationship between health stressors and psychosocial adjustment to illness remained significant but was not mediated by perception of illness impact.

Research Question: Does perception of illness impact mediate the relationship between health related hardiness and psychosocial adjustment to illness?

Hypothesis 8. Perception of illness impact will mediate the relationship between health related hardiness and psychosocial adjustment to illness; this hypothesis was supported. Perception of illness impact did mediate the relationship between health related hardiness and psychosocial adjustment to illness. Higher health related hardiness was associated with better psychosocial adjustment to illness but this relationship became non-significant when perception of illness impact was the mediator. As was noted earlier this finding supports the importance of knowing how an individual with an inherited bleeding disorder perceives the impact of the illness on his or her daily life. Pollock's (1993) also concluded that perception of the disability caused by the illness is an important variable in the process of adjusting to a chronic illness.

Research Question: Does perception of illness impact mediate the relationship between health stressors and self-perception of health status?

Hypothesis 9. Perception of illness impact will mediate the relationship between health stressors and self-perception of health; this hypothesis was not supported. The results showed no mediation of the perception of illness impact variable on the relationship between health stressors and self-perception of health status. Neither hypothesis 7 or 9 were supported. This finding would explain the lack of relationship between health stressors and self-perception of
health. The direct relationship between health stressors and self-perception of health of health status remained significant but was not mediated by perception of illness impact.

**Research Question:** Does perception of illness impact mediate the relationship between health related hardiness and self-perception of health status?

**Hypothesis 10.** Perception of illness impact will mediate the relationship between health related hardiness and self-perception of health; this hypothesis was not supported. The results did not support that perception of illness had a mediating effect on the relationship between health related hardiness and self-perception of health status. Measurement of self-perception of health status as a one-item instrument may have contributed to this result due to lower variability. Health related hardiness is an individual characteristic which promotes a feeling of control over the chronic illness, a commitment to managing this illness, and a perception of the illness being a challenge and opportunity for growth and therefore can serve as a resource to reduce the impact of the illness and improve the individual’s perception of their health. Therefore in this sample there was a direct positive relationship between health related hardiness and self-perception of health which was not mediated by the perception of illness impact. This significant relationship between health related hardiness, and greater perceived health status was also found in Pollock’s (1989) study of persons with chronic illnesses.

In summary, the significant relationship found between health related hardiness and psychosocial adaptation was a major finding of this study as depicted in Model I. This study also supported the indirect effect of perception of illness impact on health related hardiness and the psychosocial adjustment to illness. This study also provided some unexpected findings as depicted in Model I and II between health stressors, psychosocial adjustment to illness, perception of illness impact, and self-perception of health status in individuals with inherited
bleeding disorders, however replication would be worthy to strengthen the generalizability of the findings.

Limitations

This research represents the first study on psychosocial adjustment to illness in individuals with inherited bleeding disorders. Identifying limitations in design and analysis will provide for the refinement in the development of future research.

Design. The study was cross sectional so no changes in the main study variables can be determined over time.

Sample. The sample for this study was a non-random sample of convenience. From the initial potential sample of 136 participants, only 62 agreed to participate. Two were excluded from the study because their responses were received after the study was closed for recruitment. Of the participants in this study, 36.7% only had the bleeding disorder and no other chronic illness and 63.3% had more than one chronic illness. Sample size and a non-random sample were considerable limitations in this study, which limits generalizability. The power in this study was somewhat comprised because the sample consisted of 60 participants instead of the recommended sample size (64–68).

Instrumentation. The health stressor measure was a limitation in this study. The cumulative total of chronic illnesses as a measure of health stressors needs to be reconsidered in future research. The PAIS-SR had not been used in individuals with inherited bleeding disorders; therefore no normative diagnostic group was available to examine and compare the T-scores results although the diabetes group was comparable. All data was collected using self-report; disadvantages for these measures include social desirability, use of investigator developed views of the study variables and possible risk of respondent burden. Because of the sensitive nature of
the some of the PAIS domains (sexual relationships) participants may have felt that this was an invasion of their privacy.

**Strengths**

Pollock’s model provided a comprehensive framework from which to approach the study. The study models were then developed after careful consideration of the unique strengths and limitations of studying individuals with inherited bleeding disorders and a review of the prior research. All the participants in the study were followed in a specialized clinic at the Hemophilia Treatment Center of Hawaii, a state-of-the-art program with a multidisciplinary approach to care. Prior research shows utilization of hemophilia treatment centers decreases mortality within this unique population (Souice, 2000). The sample was predominantly a minority of Asian/Pacific Islander ethnicity (82%). Two of the instruments used in the study, the Health Related Hardiness Scale and the PAIS-SR both had established reliability and validity and showed reliability and validity in this study. Instruments were cost-efficient and cost-beneficial. PAIS-SR data was generated from this small sample in individuals with inherited bleeding disorders.

**Implications for Nursing Practice**

Hematological inherited bleeding disorders are difficult to identify and often obtaining an accurate diagnosis can sometimes be a complex endeavor. Appropriate referral to a federally funded hemophilia treatment center improves outcomes and leads to a more accurate diagnosis and treatment for this unique population. The findings of this study on clinical practice include:

Nurses in advanced practice need to have a good understanding of the multiple clinical problems these individuals may be facing so that they can better assist these individuals in managing their chronic illness. Encouraging individuals with inherited bleeding disorders to visit a hemophilia treatment center yearly, treating bleeds early, getting checked regularly for blood
borne infections, exercising regularly, and updating immunizations are health promotion strategies that can be incorporated into their lifestyles to live healthier lives.

Older males who had been diagnosed longer were at risk for lower psychosocial adjustment to illness in this study. In this sample female participants had more children. Referral of these patients for genetic counseling can help them to make informed decisions and obtain alternative options prior to conception. Identification of problems and meaning of findings for clinical practice can then be developed and implemented. This study showed that those who scored low on the PAIS were better adjusted to their illnesses. The study also supported the indirect effect of perception of illness impact on hardiness and the adjustment to illness. In summary, the study supported that those individuals with higher hardiness were better adjusted to their illness.

The HRHS may be a beneficial tool for use during a nursing assessment. Those found to be low in hardiness, specific interventions may be developed towards increasing the individual’s hardiness. Specific interventions such as alternative approaches in coping with stress and illness through health promotion and health prevention strategies may be implemented. Assisting the individual to manipulate his or her environment may assist him/her to have more control over it and may increase personal hardiness as well. Pollock (1989a) proposed a hardiness instruction program using small groups. As the advanced practice nurse assists individuals in increasing a sense of hardiness, it is anticipated that these strategies will assist in psychosocial adaptation and in promoting health maintenance. Enabling individuals toward successful health behaviors is an important part of nursing, and by demonstrating that hardiness is a viable construct, will assist in helping nurses meet this goal.

Nurses can utilize the research on health related hardiness to improve evidenced-based practice in caring for individuals with inherited bleeding disorders.
**Recommendations for Future Research**

Based on the findings of the research and the conclusions and implications of this study, the following recommendations are offered:

1. Replicating the present study with a larger sample size of participants with inherited bleeding disorders.
2. Improvement and refinement of the measurement scale for assessing health stressors in individuals with inherited bleeding disorders.
3. Further research using the PAIS-SR in individuals with inherited bleeding disorders to develop normative values for comparison.
4. Measure other variables such as quality of life and physiological factors and this relationship with health related hardiness and perception of illness impact.
5. Testing specific nursing interventions to promote psychosocial adjustment in individuals with inherited bleeding disorders plus other chronic illnesses.
6. Examine cultural differences in a larger sample.
7. Determine differences between carriers and different diagnosis groups with a larger sample.

**Conclusion**

In summary, this study represents the only known psychosocial research with a predominately Asian/Pacific Islander sample with inherited bleeding disorders. The results highlight the interrelationships between health stressors, health related hardiness, perception of illness impact, psychosocial adjustment to illness and self-perception of health status. However, despite the small sample size the results are meaningful in contributing to the understanding of this multifaceted concept in individuals with inherited bleeding disorders.
References


Centers for Disease Control and Prevention (1999). Estimated number of hemophilia patients in the United States. Atlanta, Georgia.


Hemophilia Data Set (2000). Data collected from federally funded hemophilia treatment centers.

Hemophilia Data Set (2002). Data collected from federally funded hemophilia treatment centers.


Holistic Nursing Practice, 13, 11-19.


Appendix A

Demographic Data Form
Demographic Data Form

1. **Age**________ (Date of Birth: __/__/__)

2. **Gender:**
   - Male___
   - Female___

3. **Ethnicity:** (Check all that apply)
   - European (Caucasian)___
   - Japanese___
   - Chinese___
   - Korean___
   - Filipino___
   - Other Asian___
   - Hawaiian or Part-Hawaiian___
   - Samoan___
   - Tongan___
   - Puerto Rican___
   - Other Hispanic___
   - African American/Black___
   - Vietnamese/Other Southeast Asian___
   - Guamanian (Chamorro)___
   - Other Pacific Islander___
   - Other (specify)__________
   - Unknown/refused________

4. **Martial Status:**
   - Married___
   - Single___
   - Divorced___
   - Widowed___
   - Separated___

5. **Education:** Fill in the highest year of education completed:
   - Elementary School___
   - Junior High___
   - High School___
   - College___
   - Graduate School___
   - Doctorate___

6. **Employment:**
   - Do you work?  Yes___  No___
   - Full-time____
   - Part-time____
7. **Occupation:** (write in)________________________________________________________________________

8. **Income:** What is your annual income?
   Under $5000 __
   $5001-10,000 __
   $10,001-20,000 __
   $20,001-35,000 __
   $35,001-$50,000 __
   $50,001-$65,000 __
   over $65,000 __

9. **Children:** Total number of children________

10. **Others in your home:** How many adults (not spouse/mate) live in your home? (write in number)
    Number of relatives______________ Number of non-relatives________

11. Have you ever been told that you have **other illnesses** such as (Check all that apply)
    ___ Asthma
    ___ High Blood Pressure
    ___ Arthritis
    ___ Diabetes
    ___ Heart Failure
    ___ Heart Disease
    ___ Hepatitis A
    ___ Hepatitis B
    ___ Hepatitis C
    ___ Cancer
    ___ Other (List)________

12. What type of bleeding disorder do you have?
    ___ Hemophilia
    ___ Hemophilia Carrier
    ___ Von Willebrand Disease
    ___ Factor V
    ___ Factor XI
    ___ Other (Please Specify)____________________________________

13. When did you find out you had this bleeding disorder?
    Month and Year____________________

**Do not put your name on this form**
Appendix B

Health Related Hardiness Scale
HEALTH-RELATED HARDINESS SCALE

Instructions:

This is a questionnaire designed to determine the way in which different people view certain important issues related to their health. Each item is a belief statement with which you may agree or disagree. Beside each statement is a scale which ranges from strongly disagree (1) to strongly agree (6). For each item we would like you to circle the number that represents the extent to which you disagree or agree with the statement. Please make sure that you answer each item and that you circle only one number per item. Thank you for taking the time to complete this questionnaire.

<table>
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1. Involvement in health promotion activities is stimulating. 1 2 3 4 5 6

2. I can avoid illness if I take care of myself. 1 2 3 4 5 6

3. I find it difficult to be enthusiastic about good health. 1 2 3 4 5 6

4. Luck plays a big part in determining how soon I will recover from an illness. 1 2 3 4 5 6

5. No matter how hard I try to maintain my health, my efforts will accomplish very little. 1 2 3 4 5 6

6. I am in control of my health. 1 2 3 4 5 6

7. I admire people who work hard to improve their health. 1 2 3 4 5 6

8. Good health is more important to me than financial security. 1 2 3 4 5 6
9. My good health is largely a matter of good fortune. 1 2 3 4 5 6

10. No matter what I do, I'm likely to get sick. 1 2 3 4 5 6

11. I find it boring to eat and exercise properly to maintain my health. 1 2 3 4 5 6

12. The main thing which affects my health is what I myself do. 1 2 3 4 5 6

13. Changes taking place in health care are not exciting to me. 1 2 3 4 5 6

14. I find people who are involved in health promotion interesting. 1 2 3 4 5 6

15. Setting goals for health is unrealistic. 1 2 3 4 5 6

16. Most things that affect my health happen to me by accident. 1 2 3 4 5 6

17. Changes taking place in health care will have no effect on me. 1 2 3 4 5 6

18. If I get sick, it is my own behavior that determines how soon I get well. 1 2 3 4 5 6

19. I do not find it interesting to learn about health. 1 2 3 4 5 6

20. I will stay healthy if it's meant to be. 1 2 3 4 5 6

21. I am not interested in exploring new ways to improve my health. 1 2 3 4 5 6
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22. No matter what I do, if I am going to get sick, I will get sick. 1 2 3 4 5 6
23. I feel no need to try to maintain my health because it makes no difference anyway. 1 2 3 4 5 6
24. The current focus on health promotion is a fad that will probably disappear. 1 2 3 4 5 6
25. No matter how hard I work to promote health for society, it never seems to improve. 1 2 3 4 5 6
26. Our society holds no worthwhile goals or values about health. 1 2 3 4 5 6
27. If I take the right actions, I can stay healthy. 1 2 3 4 5 6
28. I get excited about the possibility of improving my health. 1 2 3 4 5 6
29. I am determined to be as healthy as I can be. 1 2 3 4 5 6
30. When my health is threatened, I view it as a challenge that must be overcome. 1 2 3 4 5 6
31. I read everything I can about health. 1 2 3 4 5 6
32. I can be as healthy as I want to be. 1 2 3 4 5 6
33. When something goes wrong with my health, I do everything I can to get at the root of the problem. 1 2 3 4 5 6
34. I have little influence over my health. 1 2 3 4 5 6
Appendix C

Perception of Illness Impact Scale
Perception of Illness Impact Scale

Circle the number that corresponds to the amount your bleeding condition has affected your ability to do daily activities (Work, School) in the past twelve months.

Not Affected at all 1 2 3 4 5 6 7 8 9 10 Affected a Great Deal
Appendix D

Self-Perception of Health Status
Self-Perception of Health Status

How would you describe your overall health? (Circle one)

Poor    Fair    Good    Excellent
Appendix E

Psychosocial Adjustment to Illness Scale
**INSTRUCTIONS**

The present form contains questions concerning the effects that your illness has had on you. We are interested in knowing what effects it has had on your relationships and your ability to perform at home and on your job. Also, we would like to know about effects on family and personal relationships. Other questions concern its effects on your social and leisure time activities, and how you have felt emotionally.

In answering each question, please put a check mark (√) in the box alongside the answer that best describes your experience. Please answer all the questions and try not to skip any. If none of the answers to a question match your experience exactly, please choose the answer that comes closest to the experience you have had.

The time we would like you to report to is the past 30 days, including today. Answer each question in terms of what your experience has been like during this time period. If the event you are presently a patient in the hospital, please report your experiences for the 30 days before entering the hospital.

Some questions on the form assume that you are married or have a steady partner you are close to. Other questions ask about family relationships. If these questions do not apply to you because you are unmarried, or you have no family or partner, please leave them blank. Try to answer all the questions that do apply to you, however.

Section II asks questions about your job performance. If you have either full-time or substantial part-time employment, please answer in terms of your job. If you are primarily a student, answer in terms of your school work. If you are a housewife, answer in terms of your work at home, or any other work environment.

We appreciate the time you have taken to do this form. Please check again to make sure you have completed all the items. If you have any questions about the form, please ask. If you are responding by mail, please write them in the space provided below. Please return the form as soon as you have completed it.
SECTION 1

(1) Which of the following statements best describes your usual attitude about taking care of your health?

[ ] a) I am very concerned and pay close attention to my personal health.
[ ] b) Most of the time I pay attention to my health care needs.
[ ] c) Usually, I try to take care of health matters but sometimes I just don't get around to it.
[ ] d) Health care is something that I just don't worry too much about.

(2) Your present illness probably requires some special attention and care on your part. Would you please select the statement below that best describes your reaction.

[ ] a) I do things pretty much the way I always have done them and I don't worry or take any special considerations for my illness.
[ ] b) I try to do all the things I am supposed to do to take care of myself, but lots of times I forget or I am too tired or busy.
[ ] c) I do a pretty good job taking care of my present illness.
[ ] d) I pay close attention to all the needs of my present illness and do everything I can to take care of myself.

(3) In general, how do you feel about the quality of medical care available today and the doctors who provide it?

[ ] a) Medical care has never been better, and the doctors who give it are doing an excellent job.
[ ] b) The quality of medical care available is very good, but there are some areas that could stand improvement.
[ ] c) Medical care and doctors are just not of the same quality they once were.
[ ] d) I don't have much faith in doctors and medical care today.

(4) During your present illness you have received treatment from both doctors and medical staff. How do you feel about them and the treatment you have received from them?

[ ] a) My doctor and the medical staff have told me very little about my illness even though I have asked more than once.
[ ] b) I do have some information about my illness but I feel I would like to know more.
[ ] c) I have a pretty fair understanding about my illness and feel that if I want to know more I can always get the information.
[ ] d) I have been given a very complete picture of my illness, and my doctor and the medical staff have given me all the details I wish to have.

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From 8/10/04 To 8/9/05
(7) In an illness such as yours, people have different ideas about their treatment and what to expect from it. Please select one of the statements below which best describes what you expect about your treatment.

[ ] a) I believe my doctors and medical staff are quite able to direct my treatment and feel it is the best treatment I could receive.
[ ] b) I have trust in my doctor's direction of my treatment; however, sometimes I have doubts about it.
[ ] c) I don't like certain parts of my treatment which are very unpleasant, but my doctors tell me I should go through it anyway.
[ ] d) In many ways I think my treatment is worse than the illness, and I am not sure it is worth going through it.

(8) In an illness such as yours, patients are given different amounts of information about their treatment. Please select a statement from those below which best describes information you have been given about your treatment.

[ ] a) I have been told almost nothing about my treatment and feel left out about it.
[ ] b) I have some information about my treatment, but not as much as I would like to have.
[ ] c) My information concerning treatment is pretty complete, but there are one or two things I still want to know.
[ ] d) I feel my information concerning treatment is very complete and up-to-date.

SECTION II

(1) Has your illness interfered with your ability to do your job (schoolwork)?

[ ] a) No problems with my job
[ ] b) Some problems, but only minor ones
[ ] c) Some serious problems
[ ] d) Illness has totally prevented me from doing my job

(2) How well do you physically perform your job (studies) now?

[ ] a) Poorly
[ ] b) Not too well
[ ] c) Adequately
[ ] d) Very well

(3) During the past 30 days, have you lost time at work (school) due to your illness?

[ ] a) 3 days or less
[ ] b) 1 week
[ ] c) 2 weeks
[ ] d) More than 2 weeks

(4) Is your job (school) as important to you now as it was before your illness?

[ ] a) Little or no importance to me now
[ ] b) A lot less important
[ ] c) Slightly less important
[ ] d) Equal or greater importance than before

(5) Have you had to change your goals concerning your job (education) as a result of your illness?

[ ] a) My goals are unchanged
[ ] b) There has been a slight change in my goals
[ ] c) My goals have changed quite a bit
[ ] d) I have changed my goals completely

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- 2 -
(6) Have you noticed any increase in problems with your co-workers (students, neighbors) since your illness?

[ ] a) A great increase in problems
[ ] b) A moderate increase in problems
[ ] c) A slight increase in problems
[ ] d) None

SECTION III

(1) How would you describe your relationship with your husband or wife (partner, if not married) since your illness?

[ ] a) Good
[ ] b) Fair
[ ] c) Poor
[ ] d) Very Poor

(2) How would you describe your general relationships with the other people you live with (e.g., children, parents, aunts, etc.)?

[ ] a) Very Poor
[ ] b) Poor
[ ] c) Fair
[ ] d) Good

(3) How much has your illness interfered with your work and duties around the house?

[ ] a) Not at all
[ ] b) Slight problems, easily overcome
[ ] c) Moderate problems, not all of which can be overcome
[ ] d) Severe difficulties with household duties

(4) In those areas where your illness has caused problems with your household work, how has the family shifted duties to help you out?

[ ] a) The family has not been able to help out at all
[ ] b) The family has tried to help, but they can't do it all
[ ] c) The family has done well, and I feel somewhat relieved
[ ] d) No problem

(5) Has your illness resulted in a decrease in communication between you and members of your family?

[ ] a) No decrease in communication
[ ] b) A slight decrease in communication
[ ] c) Communication has decreased, and I feel somewhat drawn from them
[ ] d) Communication has decreased a lot, and I feel very alone

(6) Some people like yours feel they need help from other people (friends, neighbors, family, etc.) to get things done from day-to-day. Do you feel you need such help and is there anyone to provide it?

[ ] a) I really need help but seldom is anyone around to help
[ ] b) I get some help, but I can't count on it all the time
[ ] c) I don't get all the help I need all the time, but most of the time help is there when I need it
[ ] d) I don't feel I need such help, or the help I need is available from my family or friends

(7) Have you experienced any physical disability with your illness?

[ ] a) No physical disability
[ ] b) A slight physical disability
[ ] c) A moderate physical disability
[ ] d) A severe physical disability
(8) An illness such as yours can sometimes cause a drain on the family's finances; are you having any difficulties meeting the financial demands of your illness?

- a) Severe financial hardship
- b) Moderate financial problems
- c) A slight financial drain
- d) No money problems

SECTION IV

(1) Sometimes having an illness can cause problems in a relationship. Has your illness led to any problems with your husband or wife (partner, if not married)?

- a) There has been no change in our relationship
- b) We are a little less close since my illness
- c) We are definitely less close since my illness
- d) We have had serious problems or a break in our relationship since my illness

(2) Sometimes when people are ill they report a loss of interest in sexual activities. Have you experienced less sexual interest since your illness?

- a) Absolutely no sexual interest since illness
- b) A marked loss of sexual interest
- c) A slight loss of sexual interest
- d) No loss of sexual interest

(3) Illness sometimes causes a decrease in sexual activity. Have you experienced any decrease in the frequency of your sexual activities?

- a) No decrease in sexual activities
- b) Slight decrease in sexual activities
- c) Marked decrease in sexual activity
- d) Sexual activities have stopped

(4) Has there been any change in the pleasure or satisfaction you normally experience from sex?

- a) Sexual pleasure and satisfaction are still the same
- b) A marked loss of sexual pleasure or satisfaction
- c) A slight loss of sexual pleasure or satisfaction
- d) No change in sexual pleasure or satisfaction

(5) Sometimes an illness will cause a person's ability to perform sexual activities even though the person is still interested in sex. Has this happened to you, and if so, to what degree?

- a) No change in my ability to have sex
- b) Slight problems with my sexual performance
- c) Constant sexual performance problems
- d) Totally unable to perform sexually

(6) Sometimes an illness will interfere with a couple's normal sexual relationship and cause arguments or problems between them. Have you and your partner had any arguments like this, and if so, to what degree?

- a) Constant arguments
- b) Frequent arguments
- c) Some arguments
- d) No arguments
**SECTION V**

(1) Have you had as much contact as usual (either personally or by telephone) with members of your family outside your household since your illness?

- a) Contact is the same or greater since illness
- b) Contact is slightly less
- c) Contact is markedly less
- d) No contact since illness

(2) Have you remained as interested in getting together with these members of your family since your illness?

- a) Little or no interest in getting together with them
- b) Interest is a lot less than before
- c) Interest is slightly less
- d) Interest is the same or greater since illness

(3) Sometimes, when people are ill, they are forced to depend on members of the family outside their household for physical help. Do you need physical help from them, and do they supply the help you need?

- a) I need no help, or they give me all the help I need
- b) Their help is enough, except for some minor things
- c) They give me some help but not enough
- d) They give me little or no help even though I need a great deal

(4) Some people socialize a great deal with members of their family outside their immediate household. Do you do much socializing with these family members, and has your illness reduced such socializing?

- a) Socializing with them has been pretty much eliminated
- b) Socializing with them has been reduced significantly
- c) Socializing with them has been reduced somewhat
- d) Socializing with them is about the same as before, or (I have never done much socializing of this kind)

(5) In general, how have you been getting along with these members of your family recently?

- a) Good
- b) Fair
- c) Poor
- d) Very poor

(1) Are you still as interested in leisure-type activities as you were prior to your illness?

- a) Same level of interest as previously
- b) Slightly less interest than before
- c) Significantly less interest than before
- d) Little or no interest remaining

(2) How about actual participation? Are you still actively involved in doing those activities?

- a) Little or no participation at present
- b) Participation reduced significantly
- c) Participation reduced slightly
- d) Participation remains unchanged

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From 811004 To 81905

Please continue on the following page ➔
(3) Are you as interested in leisure time activities with your family (e.g., playing cards & games, taking trips, going swimming, etc.) as you were prior to your illness?

- [ ] a) Same level of interest as previously
- [ ] b) Slightly less interest than before
- [ ] c) Significantly less interest than before
- [ ] d) Little or no interest remaining

(4) Do you still participate in those activities to the same degree you once did?

- [ ] a) Little or no participation at present
- [ ] b) Participation reduced significantly
- [ ] c) Participation reduced slightly
- [ ] d) Participation remains unchanged

(5) Have you maintained your interest in social activities since your illness (e.g., social clubs, church groups, going to the movies, etc.)?

- [ ] a) Same level of interest as previously
- [ ] b) Slightly less interest than before
- [ ] c) Significantly less interest than before
- [ ] d) Little or no interest remaining

(6) How about participation? Do you still go out with your friends and do those things?

- [ ] a) Little or no participation at present
- [ ] b) Participation reduced significantly
- [ ] c) Participation reduced slightly
- [ ] d) Participation remains unchanged

(1) Recently, have you felt afraid, tense, nervous, or on edge?

- [ ] a) Not at all
- [ ] b) A little bit
- [ ] c) Quite a bit
- [ ] d) Extremely

(2) Recently, have you felt sad, depressed, or down in the stomach or of no interest in things or activities?

- [ ] a) Extremely
- [ ] b) Quite a bit
- [ ] c) A little bit
- [ ] d) Not at all

(3) Recently, have you felt angry, irritable, or had difficulty controlling your temper?

- [ ] a) Not at all
- [ ] b) A little bit
- [ ] c) Quite a bit
- [ ] d) Extremely

(4) Recently, have you blamed yourself for things, felt guilty, or lots like you have let people down?

- [ ] a) Extremely
- [ ] b) Quite a bit
- [ ] c) A little bit
- [ ] d) Not at all

(5) Recently, have you talked much about your illness or other matters?

- [ ] a) Not at all
- [ ] b) A little bit
- [ ] c) Quite a bit
- [ ] d) Extremely

(6) Recently, have you been feeling down on yourself or less valuable as a person?

- [ ] a) Extremely
- [ ] b) Quite a bit
- [ ] c) A little bit
- [ ] d) Not at all

(7) Recently, have you been concerned that your illness has caused changes in the way you look that make you less attractive?

- [ ] a) Not at all
- [ ] b) A little bit
- [ ] c) Quite a bit
- [ ] d) Extremely
Appendix F

Consent and HIPAA Form
Title of Study: Health Related Hardiness and Psychosocial Adaptation in Individuals with Inherited Bleeding Disorders & Other Chronic Illnesses.

Principal Investigator:
Mirella Vasquez Brooks, PhD Student
University of Hawaii at Manoa
School of Nursing
2528 McCarthy Mall, Webster Hall 224
Honolulu, Hawaii 96822
(808) 956-3269
(808) 956-3257 (FAX)
e-mail: vasbro96@aol.com

Faculty Sponsor:
Marilyn McCubbin, PhD, RN
Professor
University of Hawaii at Manoa
School of Nursing & Dental Hygiene
2528 McCarthy Mall, Webster 412
Honolulu, Hawaii 96822
(808) 956-5469
(808) 956-3257 (Fax)
e-mail: mccubbin@hawaii.edu

Physician Affiliation:
Desiree Medeiros, MD
Medical Director
Hemophilia & Thrombosis Treatment Center of Hawaii
1319 Punahou Street PAU
Honolulu, Hawaii 96826
(808) 983-8551
(808) 983-8005 (FAX)
e-mail: desireem@kapiolani.org

This study is being conducted as a dissertation requirement for a PhD degree at the University of Hawaii at Manoa School of Nursing and Dental Hygiene.

INTRODUCTION:
Before you decide whether or not to participate in this study, you must understand the purpose, how it may help you, any risks to you, and your rights as a research participant. This process is called informed consent. The consent form gives you information about the study and detailed instructions will also be discussed with you. It also gives you information about what health
information will be collected as part of the research study and how that information about what
health information about you will be used and disclosed. Once you understand the study, and if
you agree to take part, you will be asked to sign this consent form. If you sign this form you are
agreeing to take part in this study and to allow the research team to use and disclose your health
information. A signed copy will be given to you to keep for your reference. If you decide not to
sign this consent form, you may continue to receive care, but not as part of this research study.

Before you learn about the study, it is important that you know the following:
• Taking part in this study is of your own free will
• You may decide not to take part in the study or stop being in the study at any time. If you
decide that you don’t want to be part of this study, you will still receive medical care and
will still receive any benefits that you are normally entitled to.

It is important that the following explanation of the proposed procedures be read. It describes the
purpose, procedures, benefits, risks, discomforts and precautions of the study. It also describes
alternative procedures available and the right to withdraw from the study at any time. You have
been told that no guarantee or assurance can be made as to the results. You have also been told
that refusal to participate in this study will not influence standard treatment available to you.

You will be one of approximately 80 subjects to participate in this study.

PURPOSE:
The purpose of this study is to see how persons with bleeding disorders adjust to this condition,
how it has affected their ability to do daily activities, and how they would rate their overall
health at this time.

RECRUITMENT:
Recruitment of participants will be carried out at the Hemophilia and Thrombosis Treatment
Center of Hawaii. This outpatient setting is a federally funded clinic located within Kapiolani
Medical Center for Women & Children. The clinic nurses will identify eligible participants and
ask for permission to be contacted by the Principal Investigator (PI). The PI will obtain informed
consent. Each participant will be mailed the questionnaire after informed consent is obtained.

DURATION:
Your participation in this study will last for approximately 35 minutes.

PROCEDURES:
If you agree to take part in this study, you will be asked to complete five questionnaires. The
types of information you will be asked to provide are regarding your health. You will also be
asked questions about your illnesses. Participants will be entered into the study until the desired
enrollment goal is achieved.

FORESEEABLE RISKS OR DISCOMFORTS:
There are no foreseeable risks or discomforts associated with this study other than the time it
takes for you to participate in the study.
BENEFITS:
There will be no direct benefit from participating in this study. However, your answers and the information you provide may contribute to improving the care of persons with inherited bleeding disorders.

USE AND DISCLOSURE OF YOUR HEALTH INFORMATION:
By signing this form you are authorizing the use and disclosure of your health information. Your information will only be used/disclosed as described in this consent form and as permitted by state and federal laws.

There is no expiration date to this consent. However, if you decide to terminate your participation in this study or you are removed from this study by the principal investigator, you may revoke your consent by submitting a written notification to the principal investigator Mirella Vasquez Brooks. If you decide to revoke your consent, your health information created for this study shall not be used or disclosed by the principal investigator after the date of receipt of the written revocation except to the extent that the law allows us to continue using your information. The investigators in this study are not required to destroy or retrieve any of your health information created for this study that has been used or disclosed prior to receiving your written revocation.

Your medical records and any health information related to this study may be used or disclosed in connection with this research study to the following parties:
- Mirella Vasquez Brooks, MSN, APRN
- The Hawaii Pacific Health (HPH) Research Institution, HPH Officials, the Hawaii Pacific Health Institutional Review Board, and the HPH Office of Compliance for purposes of overseeing the research study and making sure that your ethical rights are being protected.

The individuals name above may disclose your medical records, this consent form and the information about you created by this study to:
- The faculty sponsor for this study is Marilyn McCubbin, PhD, RN
- The physician affiliation sponsor for this study is Desiree Medeiros, MD

ACCESS TO YOUR INFORMATION:
You may not be allowed to see or obtain copies of certain information in your medical or study records collected in connection with your participation in this research study while the research is in progress. Once the research is completed, you will be able to access or obtain copies of the information by submitting a written request to the Principal Investigator.

FINANCIAL COSTS TO THE SUBJECT:
There will be no financial cost to the participants.

RIGHT TO REFUSE OR WITHDRAW:
It has been explained to you that your participation is voluntary and you may refuse to participate, or may discontinue your participation AT ANY TIME, without penalty or loss of benefits to which you are otherwise entitled.
You have also been told you have the right to refuse to sign this consent. This would prevent those listed above from having access to or using any of your health information for the purpose stated. You have been told that refusal to sign this consent means that you cannot participate in this study.

OFFER TO ANSWER QUESTIONS:
This study has been explained to your satisfaction by ___________ and your questions were answered. If you have any other questions about this study, you may call Mirella V. Brooks at 956-3269.

If you have any questions about your rights as a research subject, you may call the Chairman of Hawaii Pacific Health Institutional Review Board at 535-7500 or send written correspondence to 55 Merchant Street, 27th Floor, Honolulu, Hawaii 96813 or University of Hawaii Committee on Human Studies, University of Hawaii, 2540 Maile Way, Honolulu, Hawaii 96822. Phone (808) 956-5007 or Mirella V. Brooks Principal Investigator at (808) 956-3269.

VOLUNTARY CONSENT:

I READ THE INFORMATION PROVIDED ABOVE. I VOLUNTARILY AGREE TO PARTICIPATE IN THIS STUDY AND MAY WITHDRAW MY CONSENT AT ANY TIME. MY DECISION TO WITHDRAW WILL NOT AFFECT THE MEDICAL CARE THAT I RECEIVE. I UNDERSTAND THAT MY CONSENT DOES NOT TAKE AWAY ANY OF MY LEGAL RIGHTS IN CASE OF NEGLIGENCE ON THE PART OF ANYONE WHO IS WORKING ON THIS PROJECT.

A SIGNED COPY OF THIS CONSENT FORM WILL BE GIVEN TO ME.

Subject’s Name (Print) ___________________________ Date __________

Subject’s Signature ___________________________ Date __________

Legal Representative (if applicable) ___________________________ Date __________

If signed by someone other than the subject, describe your authority to act on behalf of the subject.

________________________________________________________________________

Signature and Title of Person Obtaining Consent and Identification of Role in the Study ___________________________ Date __________
Signature of Investigator ____________________________ Date ____________________________
Hawaii Pacific Health
Authorization to Use and Disclose Protected Health Information for Research

A federal law known as the Health Insurance Portability and Accountability Act (HIPAA) sets new rules for protecting the privacy of patient’s health care records. HIPAA requires researchers, except in specific circumstances, to get written permission from study participants before using or disclosing their existing health information or new information obtained for a research study.

Purpose of this authorization:
By signing this authorization I give my permission to Mirella Vasquez Brooks and HTC and other members of the research team to use and disclose my protected health information associated with the study entitled Health Related Hardiness and Psychosocial Adaptation in Individuals with Inherited Bleeding Disorders.

Description of the information
This authorization is limited to the information that is used and/or collected about me for this study.

Who may receive my information?
I authorize the following persons or groups to receive study-related health information about me.

Expiration of the authorization
There is no expiration date to this authorization. However, I understand that if I decide to withdraw from the study or if I am removed from this study by the principal investigator. I may cancel this authorization by submitting a written notification to the principal investigator.

Canceling the authorization
I have the right to cancel this authorization at any time. My cancellation must be in writing and addressed to Mirella Vasquez Brooks. I am aware that even if I cancel this authorization and withdraw from the study, the researchers may disclose information to notify the sponsor of my cancellation, report adverse events and/or continue to use data collected if it is necessary to preserve the integrity of this study.

Disclosures outside the research study
I understand some of the persons or groups that may receive information may not be required to comply with federal privacy laws. My information may lose its federal protection if those persons or groups disclose it.

Access to Records
I understand and agree that my right to access my records that are created or obtained by ? as a part of this research may be temporarily suspended for as long as the research is in progress if the access will potentially affect the outcome of this research study. My right to access will be restored as soon as this study is completed or granting me access will not affect the outcome of this study.
Right to refuse to sign the authorization
I have the right to refuse to sign this authorization. This would prevent access to or use of my health information for this research project.
I understand that if I refuse to sign this authorization that I cannot participate in this research study. I also understand that Hemophilia & Thrombosis treatment center may not withhold or refuse to provide other treatment to me if I choose not to sign this authorization.

Written Permission
I hereby authorize the release of my health information in connection with the Health Related Hardiness and Psychosocial Adaptation in Individuals with Inherited Bleeding Disorders & Other Chronic Illnesses research study, which has been approved by the HPH Institutional Review Board (IRB). I understand that this authorization is voluntary. I will receive a signed copy of this authorization.

_________________________________________   _________________________
Signature of Research Participant            Date

_________________________________________
Printed Name of Research Participant

If signed by someone other than the participant, please describe your authority to act on behalf of the participant.

If you have any questions about HIPAA privacy protections, you may call the Hawaii Pacific Health Institutional review Board at (808) 535-7500.
Appendix G

IRB Approval Letter from University of Hawaii
MEMORANDUM

August 17, 2004

TO: Mirella Vasquez Brooks, MSN, FNP, APRN
Principal Investigator
School of Nursing

FROM: William H. Denle
Executive Secretary

SUBJECT: CHS #13131- “Health Related Hardiness and Psychosocial Adaptation in Individuals with Inherited Bleeding Disorders”

Your project identified above was reviewed and has been determined to be exempt from Department of Health and Human Services (DHHS) regulations, 45 CFR Part 46. Specifically, the authority for this exemption is section 46.101(b)(2). Your certificate of exemption (Optional Form 310) is enclosed. This certificate is your record of CHS review of this study and will be effective as of the date shown on the certificate.

An exempt status signifies that you will not be required to submit renewal applications for full Committee review as long as that portion of your project involving human subjects remains unchanged. If, during the course of your project, you intend to make changes which may significantly affect the human subjects involved, you should contact this office for guidance prior to implementing these changes.

Any unanticipated problems related to your use of human subjects in this project must be promptly reported to the CHS through this office. This is required so that the CHS can institute or update protective measures for human subjects as may be necessary. In addition, under the University’s Assurance with the U.S. Department of Health and Human Services, the University must report certain situations to the federal government. Examples of these reportable situations include deaths, injuries, adverse reactions or unforeseen risks to human subjects. These reports must be made regardless of the source funding or exempt status of your project.

University policy requires you to maintain as an essential part of your project records, any documents pertaining to the use of humans as subjects in your research. This includes any information or materials conveyed to, and received from, the subjects, as well as any executed consent forms, data and analysis results. These records must be maintained for at least three years after project completion or termination. If this is a funded project, you should be aware that these records are subject to inspection and review by authorized representatives of the University, State and Federal governments.

Please notify this office when your project is completed. We may ask that you provide information regarding your experiences with human subjects and with the CHS review process. Upon notification, we will close our files pertaining to your project. Any subsequent reactivation of the project will require a new CHS application.

Please do not hesitate to contact me if you have any questions or require assistance. I will be happy to assist you in any way I can.

Thank you for your cooperation and efforts throughout this review process. I wish you success in this endeavor.

Enclosure
Appendix H

IRB Approval Letter from Hawaii Pacific Health
August 10, 2004

Initial Approval – Expedited Review

Mirella Vasquez Brooks, MSN, FNP, APRN
University of Hawaii at Manoa
2528 McCarthy Mall, Webster 224
Honolulu, HI 96822

RE: RP #04-047-2-HPH1E
Project Title: Health-Related Hardiness and Psychosocial Adaptation in Individuals with Inherited Bleeding Disorders

Dear Ms. Brooks:

The Hawaii Pacific Health Institutional Review Board (IRB) is in receipt of your Interhospital Research Project Application, Project Summary, Informed Consent form, five (5) questionnaires, and letter of commitment from Dr. Medeiros for your study.

These materials were reviewed and approved by the IRB via expedited review on 8/10/04. The IRB has determined that the degree of risk for this protocol requires continuing review at 12 months. Therefore, this approval will expire on 8/9/05. A Continuing Review or Final Report must be submitted on or about 6/9/05.

Enclosed is your approved Informed Consent form, stamped with the dates of the approval period.

If you intend to make changes during the course of your project that will affect the human subjects involved, you must obtain IRB approval prior to implementing these changes.

Any Serious Adverse Event (SAE) or Unanticipated Adverse Event (UAE) must be reported immediately to either the IRB Chairperson or the IRB Administrative Office. A written report of the SAE or UAE must be submitted to the IRB within five calendar days. Any non-serious adverse events that may not be directly related to the study must also be reported within 45 days.

If you have any questions, please contact Joyce Fojas at 522-4544.

Sincerely,

Curtis Kamida, MD, Chair
Institutional Review Board

CK:jf

Enclosure: Approved Informed Consent form, 5 questionnaires

The Hawaii Pacific Health Institutional Review Board (IRB) operates in compliance with all applicable federal laws and regulations including but not limited to FDA regulations as described in 21 CFR parts 50 and 56, DHHS regulations as described in 45 CFR 46, guidelines resulting from the International Conference of Harmonization (ICH), the Common Rule as appropriate and operates in accordance with GCP guidelines and any applicable laws and regulations. In addition, the IRB operates in compliance with the portions of the Health Insurance Portability and Accountability Act of 1996 (HIPAA Privacy Rule) that apply to research, as described in 45 CFR Parts 160 and 164.
Appendix I

Normative Diabetic Group for PAIS-SR
Appendix F shows the Raw-scores and the T-scores for the PAIS-SR Normative values available for Diabetic patients. After consultation with the PAIS authors (Deragotis & Deragotis), diabetic patients were recommended as the comparison group. This sample consisted of 99 individuals suffering from juvenile onset, insulin dependent diabetes. All patients were participants in a multicenter national trial of an innovative therapeutic regimen designed to reduce diabetic complications through improvements in glycemic control. Approximately 36% of the sample was male and 64% female. The mean age of the group was 32.5 with little variation across centers and the group was almost exclusively white. Social class in this group followed a normal distribution across the five Hollingshead categories, with two-thirds currently or previously married and one-third single (Derogatis, 1986).
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