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THE RELATIONSHIP BETWEEN CARING FOR INDIVIDUALS DIAGNOSED WITH PRADER-WILLI SYNDROME AND CAREGIVER STRESS

A Dissertation Defense
Department of Health, Exercise Science and Recreation Management
The University of Mississippi

by

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August, 2017

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ABSTRACT

Background/Purpose: Chronic stress is related to numerous health risks such as causing an individual to age more rapidly than an individual with less stress (Oliveira, et al., 2010; Bauer, 2009; McEwen, 2002). Other aging problems related to chronic stress are Alzheimer's disease (Peterson et al., 2007) and premature death (Canizzo et al., 2011). Stress can lead to mental health issues such as depression (Wiegner, 2015; Hammen, 2005) and anxiety (Wiegner et al., 2015). Stress has also been linked to cardiovascular disease (Seldenrijk, 2015), asthma (Rod et al., 2012; Chen & Miller, 2007), obesity (McInnis et al., 2014), diabetes (Salpea, 2010), and gastrointestinal problems (Kennedy et al., 2014). The purpose of this research was to explore the relationship between caregiver stress and two primary characteristics of individuals diagnosed with Prader-Will syndrome (PWS): hyperphagia and explosive behaviors. Other variables being explored relate to variables not directly to the individual being cared for: coping strategies of the caregiver, perceived social supports, and resources/respites.

Methods: For this descriptive, cross-sectional study, an online survey was distributed by the Prader-Willi Syndrome Association (USA) to all its members between December, 2016 and March, 2017. Inclusion criteria included: participant being at least 18 years of age and not receiving pay for caring for the individual; the person being cared for being at least four years of age and living at home. A 128-item scale survey was used to assess all independent variables and their relationship with stress.

Results: A total of 278 participants completed the survey, the majority being mothers (81.3%) and Caucasian (84.2%). Most of those being cared for were female (56.1%) with the individuals being cared for having a mean age of 17.56 years of age. Cronbach's α ranged from low (α = 0.493, Self-distraction coping strategy) to high (α = 0.935, Social Provisions Scale). Multiple regression analysis found significant beta coefficients with three variables. The variable having the highest variance with stress was the Coping Strategy, Self-Blame (β = 0.257.) Social Provisions Scale (Social Supports) was the only variable which had a significant negative score (-0.182). The other variable having a significant variance was Venting (β = 0.183). The beta coefficient variance for all independent variables (R_2), including control variables was (.421). The mean score for stress was 15.96, qualifying as mild stress, with 50.7% of the participants being in the normal range.

Conclusion: The results of this study suggest that perceived social supports and two emotion-focused coping strategies have significant relationships with stress for this population. Furthermore, the variables: hyperphagia, crisis cycle of maladaptive behaviors, resources/respites, four emotion-focused and all problem-focused coping strategies had no significant relationships with stress. Findings would also suggest the participants in this study do not have the excessive stress found in other studies. Future studies using longitudinal approaches could prove beneficial to this population, as well as research investigating mediating effects of the variables identified in this study.

DEDICATION

Those who made this possible begins with my Lord, Jesus Christ. God has allowed me the time and opportunities on this earth to accomplish things needing to be done and the sense to ignore those that don't need to be done.

To the wonderful women in my life: my wife (Cathy), daughter (Jamie), and mom (Carolyn): I love you all and thank God, every day for you. Cathy, thank you for all the proofreading you did but most of all for being my biggest cheerleader during it all. You were there during those times I wanted to hang-it-up and encouraging me to continue. As with all matters in life, we were in this venture together. Jamie, your passion, diligence and fortitude in your professional career should be an inspiration to everyone. You are a credit to your profession. Mom, you were the perfect role model for anyone wanting to make a difference in others' lives. Your work ethic and love for family were instilled in all four of your sons. Your sons all know and remember the sacrifices you made for your family. Don't ever think you are not thought of, loved, and respected each day.

ACKNOWLEGEMENTS

In the realm of ideas everything depends on enthusiasm…in the real world all rests on perseverance (Johann Wolfgang von Goethe). This quote applies so well with the completion of a dissertation. I had so much enthusiasm when I was accepted into this PhD program but the enthusiasm quickly faded when all the work came due. It did not take long for the steady grind of working full time and attending classes to become a test of perseverance. With the help from others, I have finally passed that test.

I would like to thank my Dissertation Committee Chairperson, Dr. Allison Ford-Wade who has been a wonderful mentor, as well as friend. There have been numerous times your guidance has lead me through. You will be remembered for a lifetime. I also offer a special thank you to committee member and friend, Dr. Martha Bass, for all the assistance you have provided through the years. Thanks to my other committee members: Dr. Michael Dupper, and Dr. Marie Barnard for the time you put into being on my committee. I appreciate you all.

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To one of my oldest and dearest friends, Bret Johnson, his wife Michelle, and children: Lacey (Wagner), Brandt, and Brady. From the very beginning, I knew I required inspiration for such a large project. The ‘Brady Story’ provided me with such inspiration. I pray the end-results meet with your approval.

Thanks to the Prader-Willi Syndrome Association (USA): Evan Farrar, Janalee Heinemann, Sara Dwyer, and the multitude of others for their diligent work. Without your assistance, this project could not have been such as success.

And to my brothers: Mark, Joe, and Tom. You can now be reassured; pigs really do fly.

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LIST OF ABBREVIATIONS

AAIDD	American Association of Intellectual and Developmental Disabilities
ABA	Applied Behavior Analysis
APA	American Psychological Association
AS	Angelman Syndrome
ASD	Autism Spectrum Disorder
ARC	Association for Retarded Citizens
ADHD	Attention Deficit/Hyperactivity Disorder
BMD	Bone Mineral Density (BMD)
CDC	Centers for Disease Control
DASS21	Depression Anxiety Stress Scale - 21
DD	Developmental Disability
DSMR-V	Diagnostic Statistical Management Revised, Fifth Edition
FSCI	Family Stress and Coping Interview
IQ	Intelligence Quotient
IPWS	Individual with Prader-Willi Syndrome
ID	Intellectual Disability
IDD	Intellectual and Developmental Disabilities (IDD)
IRB	Institutional Review Board
PTSD	Post-Traumatic Stress syndrome

PWS	Prader-Willi syndrome
PWSA	Prader-Willi Syndrome Association
PWSA(USA)	Prader-Willi Syndrome Association: United States
SIB	Self-Injurious Behaviors
SOC	Sense of Coherence
SPSS	Statistical Package for Social Science

CHAPTER 1

INTRODUCTION

Lazarus and Folkman (1984) identified stress as a perceived burden of an event that exceeds the resources available to ensure successful management of the event. The psychological definition of stress is '*when demands from the environment challenge an individual's adaptive capacity or ability to cope*' (Cohen, Kessler, & Gordon, 1995). An organic chemist from Hungary by the name of Hans Selye coined the term '*stress*' less than eighty years ago, in 1936. Stress is considered a subjective term and Selye was not specific in his definition: "*the non-specific response of the body to any demand for change'* (The American Institute of Stress, 2015). Although the definition of stress may be subjective, the effects of stress are not.

Chronic stress is related to numerous health risks such as causing an individual to age more rapidly than an individual with less stress (Oliveira, et al., 2010; Bauer, 2009; McEwen, 2002). Stress is also related to other problems which are associated with aging such as Alzheimer's disease (Peterson et al., 2007) and premature death (Canizzo et al., 2011). Stress can lead to mental health issues such as depression (Wiegner, 2015; Hammen, 2005) and anxiety (Wiegner et al., 2015). Stress has also been linked to cardiovascular disease (Seldenrijk, 2015), asthma (Rod et al., 2012; Chen & Miller, 2007), obesity (McInnis et al., 2014), diabetes (Salpea, 2010), and gastrointestinal problems (Kennedy et al., 2014).

Parents of children with intellectual/developmental disabilities tend to exhibit chronic levels of parenting stress (Hassal & Rose 2005; Hastings & Beck, 2004; Hatton & Emerson, 2003; Head & Abbeduto, 2007; Olsson, 2008). Poor physical health (Oelofsen & Richardson, 2006) and depression (Singer, 2006) are negative outcomes associated with the caregiving of an individual with a developmental disability. Chronic parenting and/or caregiver stress is relevant to today's society because of the increasingly high prevalence rates over the years. One in six children in the United States had some form of developmental disability between the years 2006-2008. This prevalence rate increased 17.1% between 1997 and 2008 [Centers for Disease Control (CDC), 2015]. Many of these children have severe disabilities and are cared for at home by the parents in an informal caregiver role (Perrin, 2002).

Numerous studies over the past three decades have assessed factors related to the stress of caregivers of individuals with intellectual and developmental disabilities (IDD) (Hodapp, 1999; Hedov et al., 2002; Saloviita et al., 2003; Blacher & McIntyre, 2006; Lecavalier et al., 2006; McConnell & Llewellyn, 2006; Kenny & McGilloway, 2007; Khamis, 2007; Plant & Sanders, 2007; McConkey et al., 2008; Smith et al., 2008; Richman et al., 2009; Wulffaert et al., 2009; Norizan & Shamsuddin, 2010; Thomson, 2011; Pozo et al., 2014). These studies suggest that stress related health problems with this population are like the general population. Caregiver stress can be related to the behavioral, medical, and physical factors related to the specific IDD the person they are caring for may be diagnosed with (American Association on Intellectual and Developmental Disabilities [AAIDD], 2015). Although there are many studies of caregiving for individuals diagnosed with IDD and stress, very little of the research involves caregivers for individuals with Prader-Willi syndrome (PWS). High levels of stress were found by the few studies involving caregivers of individuals diagnosed with PWS (van der Borne, 1999; Thomson, 2011; Wulffaert et al., 2010; Mazaheri et al., 2013).

Prader-Willi syndrome is a genetic disorder (Driscoll et al., 2015). Griffith et al. (2011) found that parents of individuals with rare genetic syndromes are at risk for elevated stress levels and mental health problems. They conducted a study with caregivers of individuals diagnosed with three different genetic syndromes (Angelman, Cornelia de Lange, and Cri du Chat) and compared the levels of stress in those three syndromes with caregivers of individuals with autism. Those providing care for individuals diagnosed with Angelman syndrome (a genetic disorder like PWS) showed the highest levels of psychological distress among the four groups. There was a positive correlation between their maladaptive behaviors and caregiver stress. The findings were consistent with other studies showing high levels of stress in caregivers due to behavioral anomalies of certain syndromes, such as short attention span, increased sociability, hyperactivity, aggressive behavior, and sleep disorder (Clayton-Smith & Laan, 2003; Horsler & Oliver, 2006).

The goal of the present study was to provide necessary information concerning the relationship of stress in caring for an individual with PWS and the following variables: hyperphagia of individual with Prader-Willi syndrome, crisis cycle of maladaptive behaviors, fourteen coping strategies, perceived social supports, and resources/respites. The coping strategies being investigated are: self-distraction, active coping, denial, substance use, use of emotional support, use of instrumental support, behavioral disengagement, venting, positive reframing, planning, humor, acceptance, religion, and self-blame.

REVIEW OF LITERATURE

Stress

Stress occurs when a person *perceives* that he/she does not possess the needed capacity to meet the demands of the environment (Cohen et al., 1995). Lazarus (1996) also stated that stressors arise when a person perceives an inadequacy to cope with demands that threaten his/her well-being. Per the American Psychological Association (2015), there are several types of physiological stress: acute stress, episodic acute stress, and chronic stress. *Acute stress* is the most common form of stress and follows: experiencing, witnessing, or being confronted with event(s) that cause or threaten death, physical injury, or other threats to self or others (Brewin et al., 1999; Ponniah & Hollon, 2009). When acute stress is suffered too often it can result in a more severe type of stress known as episodic stress.

Episodic stress is often seen in those who self-inflict unrealistic demands, bringing too much stress in their attempt to accomplish these demands. Episodic stress can lead to Post Traumatic Stress syndrome (PTSD) which is the only major mental disorder where the cause is known: an event related to one being physically threatened (or witnessing the threat) creating intense fear, helplessness, or horror (Pitman et al., 2012). *Chronic stress* is a type of stress that is persistent and lasts a long time. Chronic stress results from long-term exposure to stressors, such as an unhappy marriage (Brock & Lawrence, 2008), traumatic experiences, unwanted career or job, poverty, and chronic illnesses (American Institute of Health, 2015). Chronic stress wears a person down and may eventually create medical and emotional issues for the caregiver (American Psychological Association [APA], 2015). Due to the long-term exposure to stress and how an individual with PWS may affect the function of the family, chronic stress will be the focus of this research project.

According to Lazarus and Cohen (1977), stressors are made by the external and internal environment; which upset the balance or homeostasis (Kenny & McGilloway, 2007). This in turn affects the physical and psychological well-being and requires action in restoring balance or equilibrium (Glanz et al., 2008). To understand how an event or events may affect a person's health, it is important to know how people may cope with stressors. Per Glanz et al. (2008), it is imperative to understand stress and coping in health education, health promotion, and disease promotion.

Coping Strategies

A person's coping skills have much to do with how well one can maneuver around life's stressors. Coping strategies are the behavioral and psychological efforts people use to master, tolerate, reduce, or minimize stressful events (John & MacArthur, 1998). Coping is also defined as a response aimed at diminishing the physical, emotional, and psychological burden that is linked to stressful life events and daily hassles (Snyder, 1999).

Coping strategies are behavioral and psychological efforts employed to overcome, tolerate, and/or reduce the impact of a stressor (Cooper et al., 2008). Research to date suggests that, while continuing to face heavy caregiving stress, caregivers can benefit greatly from structured psychosocial interventions that teach coping and problem-solving skills. (McMillan et al., 2006; Sorenson et al., 2002). Glanz et al. (2008) identified three common coping strategies people may use when faced with stressful situations; problem-focused (desire to change the stressful situation), emotion-focused (changing the way one feels about the situation), and meaning-based (attempting to induce positive emotion). Most studies have identified only the first two coping strategies, problem-focused and emotion-focused. Smith et al. (2008) claimed that emotion-focused coping strategies are less effective in reducing caregiver stress than problem-focused strategies with caregivers.

According to Carver et al. (1989), problem-focused strategies are used to attempt to alter the source of the stress. Carver goes on to claim that these strategies are used when one believes something can be done to alter the stressful situation. These strategies may include problem-solving, gathering information, weighing options, choosing between options, and acting upon choice (Holahan & Moos, 1987). Carver originally identified these strategies as: active, confrontal, planning, suppression, accommodative, restraint, positive re-interpretation and growth, seeking support, mobilizing professional help, and problem-solving (Carver et al., 1989; Hayden & Heller, 1997; Woodford, 1998; Saloviita et al., 2003; Hastings et al., 2005; Lloyd & Hastings, 2008; Glidden & Natcher, 2009, Thomson, 2011).

Emotion-focused coping strategies are used when it is believed the stressor must simply be endured (Folkman & Lazarus, 1980). This strategy focuses on reducing or managing the subjective assessment by the caregiver of the emotional effects of stress (Jones & Passey, 2004; Garland, 2007; Kenny & McGilloway, 2007; Van Der Veek, et al., 2009, Thomson, 2011). Examples of Carver's original emotion-focused strategies are: reframing or positive appraisal, positive coping, seeking social and emotional support, acceptance or passive appraisal, positive re-interpretation and growth, turning to religion, self-control, self-blame, wishful thinking, mental disengagement or distraction, behavioral disengagement, denial, focus or venting, avoidance, and managing meaning (Carver et al., 1989, Kramer, 1993; Hayden & Heller, 1997; Grant & Whittell, 2000; Saloviita et al., 2003; Hastings et al., 2005; Lloyd & Hastings, 2008).

Measuring how problem-focused and emotion-focused coping strategies differ in their effectiveness has been problematic. Carver (1989) found such a problem with the definition of coping strategies between authors. One author may identify a coping strategy as emotional-focused, while another may label it as problem-focused. Carver’s own 60-item COPE instrument also has difficulty distinguishing between emotion-focused and problem-focused strategies. Another issue with the COPE is the time constraints of research participants in completing the instrument. A newer instrument, the Brief COPE was created with 14 scales and 28-items, two items per scale. This measurement instrument greatly shortens the amount of time it takes to finish the survey (Carver, 1997).

The newer, briefer instrument provides a total of 14 dimensions of coping strategies and is used to measure types of coping strategies individuals use in dealing with stress (Carver, 1997). Per Tuncay et al. (2008) the Brief COPE has a total of 28-items of which two questions are asked for each of the 14 subscales for coping strategies. It is a self-reporting instrument that measures problem-focused and emotional-focused coping skills. The problem-focused strategies of the Brief COPE are not the same as in the original COPE. Those problem-focused skills in the Brief COPE are: acceptance, religion, planning, positive reframing, use of instrumental support, active coping, use of emotional support, and humor. The emotion-focused coping strategies for the Brief COPE are: self-distraction, venting, self-blame, behavioral disengagement, denial, and substance use (Carver, 1997; Tuncay et al., 2008; Yusoff et al., 2010).

Hastings et al. (2005), conducted factor analysis on the Brief COPE and extracted four coping strategies: active avoidance (emotion-focused), problem-focused (problem-focused), positive coping (problem-focused), and religious/denial coping (emotion-focused). The study was conducted with mothers and fathers of children with autism. Active avoidance identified active attempts to avoid the stressor or escape from its effects. This factor contained the following elements of the Brief COPE: substance use, behavioral disengagement, self-blame, venting of emotions, and distraction. Problem-focused, as the title states, represents the problem-focused strategies within the Brief Cope: planning, active coping, seeking instrumental support, and seeking emotional social support. Positive coping is best described as attempting to adopt positive coping strategies and contained the following elements of the Brief COPE: humor and positive reframing, acceptance, and emotional social support. The fourth factor, religious/denial included all the elements from religious coping and denial from the Brief COPE. Carver (1997) claimed that one concern with many coping scales is that authors categorize some strategies as problem-focused while others may view them as emotion-focused. Others may view some emotion-focused as problem-focused, Carver stated that his scales are no different.

Heath Risks of Stress

There is a significant relationship between stress and disease. Chronic stressors and responses to them affect the sympathetic nervous system and endocrine functions, thus influencing the occurrence and progression of health problems such as cancer, infectious disease, and HIV (Glaser & Kiecolt-Glaser, 2005; Glanz, et al., 2008). Cortisol is a hormone closely associated with stress. Plasma and salivary cortisol are used as an index of cortisol hypothalamic-pituitary-adrenal axis activity when emotional distress is widespread (Melamed et al., 1999; Buchanan et al., 1999). Stress increases levels of cortisol which elevates risk of disease (Esch, 2002a; Vedhara et al., 2003).

Bauer et al. (2009) stated that aging (senescence) of the immune system, Immunosenescence, is a disorder of the immune system involving age and is closely related to chronic stress and other stress factors. There is a relationship between chronic stress, caregiving, and the shortening of telomeres. Telomeres are DNA-protein complexes that cap the ends of chromosomes and promote chromosomal stability. The effect on telomeres by chronic stress will eventually lead to premature senescence (aging) as well as onset of disease, including cardiovascular disease (Epel et al., 2004). Telomere length is related to elevated stress hormones (catecholamines and cortisol). Low telomerase activity is associated with the major risk factors for CVD, such as: smoking, poor lipid profile, high systolic blood pressure, high fasting glucose, and greater abdominal adiposity (Epel et al., 2004). Stressors play a major role in immunological diseases and immune-related disease processes. These stressors may eventually create inflammation, infection, autoimmune processes, and even malignant tumors (Esch et al., 2002b).

Intellectual and Developmental Disabilities

Intellectual functioning or intelligence references a person's general mental capacity in areas such as learning, reasoning, and problem solving. The Intelligence Quotient (IQ) test is one way to measure intellectual functioning. An IQ around 70 indicates there is some form of limitation in this area (AAIDD, 2015). The ceiling for this threshold can go as high as 75 when taking into consideration the standard error of measurement of approximately five. This score is approximately two standard deviations below the mean (AAIDD, 2008). One must remember that an IQ of 70 is only the threshold of being diagnosed with an intellectual disability. Another issue to determine if a person has an intellectual disability is adaptive skills.

AAIDD (2015) identifies adaptive behavior (skills) as the collection of everyday living skills learned and performed by people in the areas of conceptualizing, socializing, and practical skills. Limitations in these adaptive behaviors can be determined by standardized tests:

- Conceptual skills are in the areas of language and literacy; money, time, number concepts, and self-direction (Tasse et al., 2012).
- Social skills are interpersonal skills, social responsibility, self-esteem, gullibility, naïveté (i.e., wariness), social problem solving, ability to follow rules, obey laws, and to avoid being victimized (Tasse et al., 2012).
- Practical skills are activities of daily living (personal care and hygiene), occupational, healthcare, travel/transportation, schedules/routines, safety, use of money, and the use of the telephone (Tasse et al., 2012).

Basic terminology used in identifying those diagnosed with one of many IDDs can be confusing, let alone any discussion of the anomalies associated with any one IDD and caregiver stress associated with that IDD. One problem is that many studies interchange the terms Intellectual Disability (ID) and Developmental Disability (DD) when identifying the population being studied. These two terms are not synonymous. In fact, a person diagnosed with a DD does not necessarily have a diagnosis of an ID (Smith, 2010). Yet, this confusion can be partially explained by the definition and classification system for IDD changing three times in the past two decades (Intellectual Disabilities Definition, 2011; American Psychiatric Association, 2013).

The global prevalence rates for ID range from 1% - 3% (Harris, 2006). Per a meta-analysis conducted by Maulik et al. (2011) which included 52 studies, the global prevalence rate of ID is estimated at 10.37/1000. This would be consistent with United States and other developed countries' prevalence rates provided by Harris (2006) and King et al. (2009). One must meet three criteria to be diagnosed with an ID; a three-prong test. Those criteria are in the areas of: 1) IQ scores, 2) adaptive skills, and 3) age at time of diagnoses (AAIDD, 2015).

The AAIDD defines an intellectual disability as: a disability characterized by significant limitations in both intellectual functioning (first prong) and inadaptive behavior (second prong), which covers many everyday social and practical skills. This disability originates before the age of 18 (third prong) and is a life-long disability, requiring caregivers to ensure the welfare of the individual from birth well into their adulthood (AAIDD, 2015).

King et al. (2009) states that the ID prevalence rate could be as little as 1% when factoring in that some of these cases could be: diagnosed as a learning disability (a different form of developmental disability); the result of a deficit in adaptive skill interfering with testing; issues with varying rates of cased identification, and mortality (King et al., 2009). Many factors are considered when estimating prevalence rates by researchers. Maulik (2010) lists these factors as: diagnostic criteria, severity of illness, gender, age, study population, and socio-economic status. Prevalence rates for intellectual disabilities vary greatly between ages as well: Beange & Taplin (1996) in an Australian study estimated the prevalence rate for 20 – 50 year olds at 3.3/1000. Leonard et al. (2003) provided an estimate of 14.3/1000 in a study of 6 to 15-year-old Australians (Maulik, 2010).

The term developmental disability (DD) is often used in conjunction with ID but in fact is an umbrella term which may include numerous other disabilities (The Arc, 2015). The diagnosis of DD can include disabilities that are apparent during childhood, but can manifest well after teenage years. Unlike the diagnosis of an ID being required by the age of 18; one can be diagnosed with a DD prior to his/her 22nd birthday (AAIDD, 2015). Like an ID, DDs may also be lifelong, severe, and chronic. Developmental disabilities are mostly physical, yet some conditions include an intellectual disability. Examples of this would be Down syndrome or fetal alcohol syndrome, both of which have been studied thoroughly (AAIDD, 2015; ARC, 2015).

Intellectual disability is the most common developmental disability, having an immense effect on the individual, his/her family, and even the community (Maulik & Harbour, 2010). For children under the age of 15, prevalence is higher among males than females. This could be a result of males displaying more abnormal behaviors in school compared to females (Maulik & Harbour, 2010). Since ID is a subcategory of DD; one would understand prevalence rates for DD being much higher than ID. Boyle et al. (2011) claimed the prevalence rate for children with DD in the United States between the years of 1997–2008 was identified as 15.04%; with one in six being diagnosed with a DD. A study conducted in 2008 found nearly 10 million children in the US had a diagnosis of DD, a 12% increase in 12 years (Pettapiece, 2007). There were 1.8 million more children with DD during 2006-2008 than during the prior decade. Some of those significant increases were in the areas of autism (289.5% increase), Attention Deficit Disorder (33% increase), and hearing loss (30.9%) (CDC, 2014). These disabilities have received much attention in modern research. One specific disability lacking research is Prader-Willi syndrome (PWS), especially with caregiver stress.

To explain what IDD is, one must identify and explain the diagnoses with IDD. This is a term used in diagnosing intellectual disabilities as well as those being diagnosed with developmental disabilities. One must distinguish that an individual who may have a diagnosis of ID may also have another diagnosis of DD. However, a person with a diagnosis of DD may not necessarily have a diagnosis of ID (Jasien et al., 2012). In fact, ‘Developmental Disabilities’ is an umbrella term which includes numerous specific disabilities. One of the disabilities falling under this umbrella term is ‘Intellectual Disabilities’. Within the literature, the terms DD and ID are often used interchangeably; often being referred to as IDD. Throughout this dissertation, the terms will often be used in conjunction with each other (IDD) but will be broken apart at other times (ID and/or DD).

To further explain the relationship between ID and DD, one must define both separately. Developmental disabilities is used to identify anyone who may have one or more of the following disabilities: breathing, vision and/or hearing, bone or joint, injuries (including traumatic brain injury), epilepsy or seizures, speech, learning disabilities, attention deficit hyperactivity disorder (ADHD), intellectual disabilities (ID), mental, emotional, behavioral, and other types of birth defects (Houtrow et al., 2014) including cerebral palsy (Bax et al., 2005), autism or other neurological conditions (Elsabbaghm et al., 2012). Developmental Disabilities are life-long, pervasive conditions that may negatively affect individuals’ cognition as well as health. These conditions must impair general intellectual functioning or adaptive behaviors. A person must be diagnosed with these impairments prior to turning 22 years of age to be diagnosed with a developmental disability (CDC, 2014).

Adaptive behaviors must also be two standard deviations below the mean and must be in at least two of ten areas (AAIDD, 2008). These areas are: communication, self-help, home living, social skills, community use, self-direction, health and safety, functional academics, leisure, and work. Both IQ and adaptive skills must be measured by a standardized instrument. (Developmental Disabilities Resource Center, 2014).

As stated above, ID is listed as one of the subcategories of DD. There is a three-prong test that must be met to be diagnosed with an ID. In 2011, The American Association on Intellectual and Developmental Disabilities (AAIDD) stated that an individual must have a significant limitation in both intellectual functioning and in adaptive behaviors in conceptual, social, and practical skills to be diagnosed with ID. The diagnosis must also be provided prior to the age of 18. AAIDD's definition allows a person who may not be diagnosed with an ID at the age of 18 to still be diagnosed with another form of DD before the age of 22.

To have a diagnosis of ID requires the person's intelligent quotient (IQ) to be a minimum of two standard deviations below the mean. The mean IQ is 100 with a standard deviation of 15 (AAIDD, 2008). This would require an individual to have an IQ of less than 70 to be considered as having an impaired intellectual functioning. The Diagnostic Statistical Management Revised, Fifth Edition (DSMR-V) identifies four classifications for ID: Mild (IQ 69-55), Moderate (IQ 54-40), Severe (IQ 39-25), and Profound. (IQ<25) (Jasien et al., 2012).

Developmental disabilities encapsulate both those who have been diagnosed with an ID by the age of 18 and/or diagnosed with a DD before they turn 22 years of age. The health of individuals with IDD is of vital importance considering the prevalence rates and multiple chronic health conditions these people may acquire. Beange (1996) claimed that most individuals with IDD are not "sick" nor have "ill health" and listed three major factors that determine individuals with IDD health risks: genetic composition, lifestyle, and the increase of lifespan. Rubin & Crocker (2006) listed low bone mineral density (BMD) and osteoporosis as one of the health issues facing individuals with IDD (Goldstone et al., 2008; Vice et al., 2015). Other health issues facing this population are: thyroid disease, cardiovascular problems, respiratory infections, gastroesophageal reflux disease, dental and oral hygiene issues, and behavior problems. Poor nutrition and sedentary lifestyles are also major health concerns for those being diagnosed with IDD (Rimmer & Yamaki, 2006).

Intellectual and Developmental Disabilities and Stress

There have been a significant number of studies identifying stress as an unhealthy outcome of the caregiving of individuals diagnosed with IDD. These stressors vary as both the individual with IDD (Minnes et al., 2007) and the caregiver ages (Hogg et al., 2001). It is important for researchers to address stress related factors in hopes to educate and prevent dangerous stress levels among caregivers. In addition, providing healthcare service providers and policy makers with the necessary knowledge about the dangers of stress among this population is crucial. Doing so would provide service providers and policy makers the necessary knowledge so they can provide support and resources/respites needed for both the individual as well as their caregiver. Per Son et al. (2007), higher levels of subjective as well as objective stressors in caregivers are associated with; self-reported poorer caregiver health; more negative health behaviors; and greater use of health care services. Objective health can be mediated by the feeling of overload by the caregiver.

Caregivers of individuals with IDD have also been found to have higher levels of stress compared to the general population. Spousal support has shown to have an affect on distress levels of primary caregivers. One study found a negative relationship between primary caregiver stress of cognitively impaired elders and emotional support provided by a secondary caregiver. When the same secondary caregivers provided instrumental support along with emotional support to the primary caregivers, there was a likelihood of lower psychological distress of the primary caregiver when the one being cared for had symptoms of a greater negative mood (Lou et al., 2013). This would suggest that emotional support provides psychological relief to the primary caregiver but they may need more professional and experienced assistance with the day-to-day responsibilities of caring for an individual.

The severity of an individual's symptoms of a disability could affect the level of distress a caregiver may display. Benson (2006), found a significant positive correlation between severity of symptoms of individuals with autism (*n*=61) and their parents' (*n*=68) depression and stress proliferation (Benson, 2006). This will specifically affect the mothers' perception of her family's quality of life more than the fathers, possibly related to their experience as primary caregivers and to their higher degree of responsibility for parenting (Pozo et al., 2014).

The effects of caring for an individual with an IDD with behavioral problems can be extensive, restrictive, as well as disruptive to the family. It can affect the family economically, socially, as well as emotionally (Khamis, 2007). Behaviors such as aggression and self-injurious behaviors (SIB) have shown to be the biggest contributor to caregiver stress in Autism (Donenburg & Baker, 1993; Tominik et al., 2004; Lecavalier et al., 2006; and Pozo et al., 2014). One may think that maladaptive behavior and caregiver stress is one directional; behavior is always the predicting variable and stress the dependent variable. On the contrary, maladaptive behaviors and stress have been shown to exacerbate each other (Lecavalier, Leone, & Wiltz, 2006) suggesting that if a caregiver can control stress, it may have a positive affect on the behavior of the individual. When behavior problems are more severe, parents may perceive the situation as less predictable, less manageable, and less meaningful, causing more stress (Pozo et al., 2014).

Studies suggest that predictors of caregiver and family stress include the age of the individual (Khamis, 2007). This stress would begin almost immediately upon the birth of that person. McConkey et al. (2008) claimed that almost immediately there are three stressors the caregiver must face upon the birth of an individual with a disability: the diagnostic process, the realization that the child will not have a ‘normal life’, and the feeling of grief and guilt. This stress may lessen once the cause of the child’s disability is explained (Lenhard et al., 2005; Skotko, 2005; Graungaard & Skov, 2007; and Thomson, 2011) and proper information about the disability is provided to the caregivers (Skotko, 2005; Graungaard & Skov, 2007; Kenny & McGilloway, 2007; and Thomson, 2011). This would further imply that professionals and medical teams need to be knowledgeable of diagnoses, treatment planning, and resources available to caregivers of individuals in lessening their stress.

The characteristics of children are not the only factors influencing the stress of caregivers (Goldstone, 2008). Being able to adapt to the issues surrounding their children’s disability is another factor (Pozo, 2014). Many factors have been demonstrated to be significantly related to adaptation processes. Many families can adapt well and appear to thwart-off the stressful effects of raising a child with severe IDD and behavioral problems (Gerstein et al., 2009). Personal and family resources can be good predictors of caregiver stress as well. Adequate resources to enable meeting the demands of the individual are very important in successfully adapting and avoiding stress for caregivers and families of IDD (Khamis, 2007). These resources are often lacking, as well as the services being offered to the caregivers (Maes et al., 2003).

Prader-Willi Syndrome:

Prader-Willi syndrome is a genetic disorder characterized by several anomalies such as: hypotonia and obesity (Driscoll et al., 2015). This genetic disorder was originally named after two of the three medical doctors who formally diagnosed the disorder ‘Prader-Labhart-Willi’ in 1956 (Prader et al., 1956; van den Borne, 1999; Panich, 2003). Andrea Prader was a Swiss pediatrician and endocrinologist (Enerson, 2001; Panich, 2003). Alexis Labhart was originally from Russia, eventually living in America and graduating from Harvard with a background in internal medicine, tuberculosis, and endocrinology (Beighton, 1986; Panich, 2003). Heinrich Willi was a Swiss pediatrician specializing in endocrinology and neonatology (Breighton, 1997; Panich, 2003).

Prader, Labhart, and Willi found common variables among nine children with developmental disabilities and went on to conduct further research in PWS. Their research associated several anomalies to this disability, one of which is hypotonia (floppy-baby) during infancy (van der Borne, 1999). Per Driscoll et al. (2014), there are behavioral phenotypes that are very common in PWS as well; temper tantrums, stubbornness, manipulative behaviors, and obsessive-compulsive characteristics. Phenotypes are observable, physical, and/or biochemical characteristics of the expression of a gene.

The list of major anomalies that may lead one to be diagnosed with PWS are: infantile hypotonia; numerous cranial facial features; developmental disabilities; hypogonadism; feeding problems and failure to thrive in infants; and rapid weight gain for young children (Buiting, 2010; van der Borne, 1999; Holm et al., 1993). There are less significant anomalies of PWS that may not be life threatening but are issues of concern when assessing stress in caregivers. These may include: decreased fetal movement and lethargy in infants; thick saliva; hypopigmentation; short stature; Estropia/Myopia; narrow hands/feet; defects in articulation; sleep disturbance; apnea; and psychiatric issues (Holm, 1990; van der Borne, 1999; Schieman, 2003; Cassidy and Driscoll, 2009; Mazaheri et al., 2013; Driscoll, 2014). Two of the psychiatric issues will be addressed as stressors in this research project.

Other anomalies that may create stress for parents/caregivers would include: decreased vomiting (less gag reflex) which creates concerns for overeating; high pain threshold; low bone mineral density; and other orthopedic and bone abnormalities such as osteoporosis and scoliosis (Holm, 1990; Schieman, 2003; Panich, 2003; Driscoll, 2009; Vice et al., 2015; Jasien et al., 2012; Mazaheri et al., 2013; Goldstone, 2013). Although PWS was formally identified in 1956, there is documentation, literature, and archeological finds suggesting that others have observed and/or been conducting research with individuals with similar characteristics as early as 4,000 BC (Panich, 2003).

Many individuals diagnosed with PWS have cognition problems (van der Borne, 1999; Driskoll et al., 2015; Dykens et al., 2000; Dykens & Shah, 2003; Buiting, 2010). Most individuals diagnosed with PWS score in the borderline, mild, or moderate range of IQ with their adaptive functioning appearing lower than their IQ would suggest (Dykens, 2000). Regardless of the severity of the cognitive issues, most children with PWS have multiple severe learning disabilities and poor academic performance (Whittington et al., 2004). This genetic disorder is related to the 15th chromosome and often affects an individual's cognition (Intellectual Disability) while posing health problems associated with being diagnosed with a DD (Driscoll et al., 2015). Prevalence rates for PWS in the 1990's were between 1 in 16,062 (Burd et al., 1990) and 1 in 25,000 (Butler et al., 1990). As late as 2015, prevalence rates have been estimated between 1 in 12,000 and 15,000 (Prader-Willi Syndrome Association, 2015). Those prevalence rates have not changed much in the past 25 years. Cassidy and Driscoll (2009) list prevalence rates of PWS at 1/15,000 to 1/30,000 within the United States. Prader-Willi syndrome occurs in males and females equally (Butler et al., 2006) and in all races (Prader-Willi Syndrome Association, 2015). Prader-Willi syndrome is the most common syndromic form of obesity affecting between 350,000 and 400,000 individuals worldwide (Butler et al., 1990; Whittington et al., 2001; Vogels et al., 2004).

Much like caregivers of other IDDs, those caring for individuals with PWS can be at risk for negative health outcomes due to the stress related to caring for their loved one. Yet, there are numerous medical, physical, and behavioral anomalies that often coincide with the cognitive concerns of PWS (Goldstone, 2013). Other issues can create additional stress for caregivers that are not often associated with other disabilities. Medical concerns for individuals with PWS include: low muscle tone, short stature (if not treated with growth hormone), and incomplete sexual development. One major medical concern associated with PWS is the chronic and life threatening health problems associated with hyperphagia, or excessive eating. Hyperphagia is the result of insufficient functioning of the hypothalamus which requires the individuals to be placed on strict diets (Dykens et al., 2000; Goldstone et al., 2004).

Historically, practitioners believed that individuals with PWS had two phases of eating issues: *failure to thrive* and *hyperphagia* leading to obesity, [(Wulffaert et al., 2009; Prader-Willi Syndrome Association (PWSA), 2015)]. Another study (Miller et al., 2011) and report (Goldstone, 2008) found transitioning between nutritional phases to be much more complex, with seven different nutritional phases through which individuals with PWS typically progress; from infancy to adulthood. Refer to Table 1 for the nutritional phases.

Table 1. Nutritional Phases in PWS

Phases	Median Age	Clinical Characteristics
0	Prenatal-birth	Decreased fetal movements and lower birth weight
1a	0-9 months	Hypotonia with difficulty feeding and decreased appetite
1b	9-25 months	Improved feeding and appetite: growing appropriately
2a	2.1-4.5 years	Weight increasing without appetite increase or excess calories
2b	4.5-8 years	Increased appetite and calories, but can feel full
3	8 years-adulthood	Hyperphagic, rarely feels full
4	Adulthood	Appetite is no longer insatiable for some

Miller et, al. (2011)

This insatiable appetite coupled with a metabolism that utilizes drastically fewer calories than normal can later result in obesity and health problems related to obesity (i.e. diabetes and heart disease) (Butler et al., 2002). The reduced physical activity of these individuals (van Mil et al., 2000) compounds this problem. Caregivers of individuals with PWS often must restructure their lives around these serious and life threatening medical concerns. It requires caregivers to limit access to food by placing their loved ones on strict diets increasing the monitoring of food, as well as locking food away from the individual (food seeking behaviors) (Goldstone, 2008). Because of this *regulation* of food to prevent future medical problems; immediate behavioral problems may very well occur. Examples of these behavior problems are; aggression, food seeking, tantrumming, pica (eating foods considered inedible), narcissism, manipulation to obtain more food, and running away. These maladaptive behaviors may be displayed individually or in combination while attempting to access food. These behaviors, such as hoarding and skin picking, along with other factors associated with having a child with a genetic syndrome and/or IDD places caregivers at great risk for elevated levels of stress (Griffith et al., 2011).

Individuals with PWS may require care throughout their life while those caring for them encounter specific and significant disabilities that are age-related. These disabilities account for high stress levels reported by caregivers (van den Bourne, 1999; Thomson, 2011; Mazaheri et al., 2013).

Stress for caregivers of individuals with PWS may begin almost immediately. The realization that one's newborn child is severely disabled would be stressful (Zolnit & Stark, 1961; Blacher, 1984; Qhine & Pahl, 1987; Beresford, 1994; Case, 2001; Graungaard & Skov, 2007). The diagnostic process can create an emotional and traumatic experience for the caregivers of the individual (Hogg et al., 2011; Cantrell-Bartl, 2006). Caregivers may be unable to seek employment due to being required to stay home and care for their loved one. This may further present stressful environments due to financial hardships, as well as creating stress related to spending extended periods of time alone handling the day-to-day concerns that exist with individuals diagnosed with PWS (Treadwell et al., 1995; Goldstone et al., 2008).

As individuals diagnosed with PWS age, their needs may change. The needs of a newborn child may not be as relevant as proper and adequate education during the individual's childhood or teenage years. Schools are often not set up to facilitate the educational needs of these children. Often a parent must accompany the child to school to intervene and assist with many of the activities offered. This would prevent that parent from engaging in suitable employment. Recreational and leisure opportunities offered by after-school programs and other non-school related activities may be difficult to attend as well. Behaviors mentioned earlier may interfere with these children being accepted by other children, or even the possibility of not being allowed in the programs.

As the child matures into adulthood, other sets of issues present themselves that they and the caregivers must struggle with (Schrander-Stumpel et al., 2007). When most caregivers are experiencing their children moving onto college or a career; the caregivers of these individuals must maneuver around a system of Bureaucracy attempting to find meaningful employment and adequate housing for the individual they are caring for (AAIDD, 2015). Again, this would create difficulty in achieving meaningful careers for the caregivers themselves.

As the child ages, so does the caregiver. There are serious questions caregivers must face as they face mortality: Who will care for their loved one and where will he/she live? Again, these uncertainties create stress as they have throughout the individual's life. Regardless of the age of the individual with PWS; resources, supports, and information are needed to assist in the care for an individual.

The lack of a comprehensive community, long-term support services in the US for individuals diagnosed with intellectual and/or developmental disabilities is at a crisis level (AAIDD, 2015). Research suggests that stress for caregivers of individuals diagnosed with PWS is more intense than for those caring for individuals with other developmental disabilities and that proper supports and resources are beneficial in lessening caregiver stress (van den Borne et al., 1999; Thomson, 2011, and Mazaheri et al., 2013). Wulffaert et al. (2009) also found mothers of individuals with PWS to have high levels of stress.

Hastings & Beck (2004) suggest the provision of parental support when a child exhibits substantial behavioral problems as reducing stress. Hastings & Beck (2004) also emphasize the need for caregivers to have access to adequate information concerning the disability. Murphy et al., (2006) suggests greater knowledge of caregiver health-related needs to allow for the improvement of existing services, as well as developing new strategies would better assist caregivers in their vital roles.

Social supports have been shown to boost coping skills for families of children with emotional and behavioral challenges (McDonald et al., 1997; McDonald et al., 1999; Brannan et al., 2001). If support, as well as information is provided to caregivers concurrently, caregiver satisfaction may be improved (Wulffaert, 2009). Wulffaert (2009) also suggests that future studies include fathers to ascertain perceptions of how their child's behavior affects them. Furthermore, the present study will provide organizational leaders and policymakers additional information to solicit needed funding to offer supports and resources for these caregivers. Results will be available for later interventions addressing coping skills that may be beneficial in handling hyperphagia and the crisis cycle of maladaptive behaviors for caregivers for individuals diagnosed with PWS. Unlike previous studies conducted with this population, the database from the PWS (USA) will be used to solicit a large enough sample size to better generalize to others caring for those diagnosed with PWS within the US.

Research concerning relationships between stress and the caregiving of individuals with PWS is limited. Most research on caregiver stress has been conducted with other forms of IDDs. Those disabilities include: i*ntellectual disabilities* Khamis (2007), McConkey (2008); *Autism*

Lecavalier et al. (2006), Smith et al. (2008), Pozo et al. (2014), Weiss & Lunsky (2011); *learning disabilities* (Kenny & McGilloway, 2007); and *multiple diagnoses* Plant & Sanders (2007), Blacher, & McIntyre (2006).

Intellectual/Developmental Disabilities (Non-PWS Related) Stress Studies

Lecavalier et al. (2006) conducted a study with parents and teachers of 293 individuals diagnosed with autism. Findings of the study for the parent evaluation portion of the study suggested that maladaptive behaviors displayed by the individual had the largest association with caregiver stress than any other characteristic. The study found no large association between caregiver age, education level, or knowledge of Applied Behavior Analysis in autism (ABA: a tool used in effectively working with individuals who display maladaptive behaviors) and stress. The individuals' age and gender had no significant relationship with caregiver stress either. Specifically, conduct problems and poor prosocial behaviors of the individual had the strongest association with caregiver stress. These findings suggest that individuals who were disruptive or broke rules created more stress for caregivers than individuals who did not display these behaviors. Another result of this study was that maladaptive behaviors and stress exacerbated each other, suggesting that addressing ways to reduce stress in the caregiver could have a positive effect on the individual's behavior (Lecavalier et al., 2006).

Blacher & McIntyre (2006) interviewed caregivers (*n*=282) of individuals diagnosed with ID. Participants were primarily mothers but also included adoptive mothers, stepmothers, or grandmothers (all ranging from 31-70 years of age). The study examined how behaviors of individuals between the ages of 16-26 of low functioning level (with differing IDs) affected the stress of their caregivers. The Axis II diagnoses of the individuals were: Down syndrome (*n*=59), autism (*n*=23), cerebral palsy (*n*=87), and undifferentiated (*n*=113) and ranged from moderate to severe ID. The caregivers included Southern Californian Anglo participants (*n*=150) and Latinos (*n*=132), who spoke primarily Spanish.

In the study by Blacher & McIntyre (2006), autism proved to have the highest levels of behavioral problems among individuals. This group also showed the lowest level of well-being among mothers. Individuals diagnosed with Down syndrome showed the lowest level of maladaptive behaviors as well as highest well-being among their mothers. When behavior was controlled for, there were no differences between maternal stress and depression by individual Axis II diagnostic grouping. There were no pattern differences between Anglos and Latinos in the areas of maladaptive behaviors as well as in maternal well-being. The level of well-being did change however. Latino mothers reportedly had lower levels of morale and higher levels of depression than did their Anglo counterparts. The Latino mothers also showed higher levels of positive impact from their child than did the Anglo participants. This study showed that behavioral issues had a positive correlation with caregiver stress.

Difficulty in tending to tasks associated with IDD along with maladaptive behaviors plays an important role in caregiver stress. Plant & Sanders (2007) studied mothers (*n*=105) and fathers (*n*=34) of children with various levels of severity IDD living in Queensland, Australia under the age of six. The children had diagnoses of autism, cerebral palsy, Down’s syndrome, and chromosomal disorders other than Down’s syndrome. The study evaluated the predictors, mediators, and moderators of parental stress and found maladaptive behaviors of children to be second only to difficulty levels of the parental caregiving tasks for the child.

A tool designed by Shearn & Todd (1997) was used to assess tasks performed by parents, and the Revised Family Observation Scale was used to measure negative behaviors of the children. Issues associated with food, including maladaptive behaviors, were found most stressful for parents in the Plant & Sanders (2007) study. Assisting with mealtime was the best predictor of parental stress. Other predictors of parental stress were cleaning up after the child; settling down at bedtime, as well as getting ready for bed. Various aspects of maneuvering around local resources were listed as good predictors of parental stress as well (advocating for the child; transportation, medical appointments, reading information related to the disability, filling out forms, and attending parental training/meetings/seminars). Preparing special meals was another area of concern with parental stress in this study.

Murphy et al. (2006) studied families caring for children with a multitude of disabilities. This study of 40 caregivers included mothers (*n*=33), fathers (*n*=6), and one aunt/legal guardian. Participants listed lack of day-to-day control over events as a major stressor as well as the need to continuously advocate for their child. Concerning health changes in the past year: 16 (40%) reported a worsening of health, 20 (50%) reported no change, and four (10%) felt their health had improved. Many of these caregivers claimed that meeting the day-to-day needs of their child and concerns of the future created considerable stress, twenty of which had received some form of intervention by a mental health provider.

Coping skills used in this study consisted of: frequent breaks, mini-naps, crying, pets, shopping, and eating chocolate. Many stated that speaking to friends by telephone was effective in reducing stress. Many of these caregivers used informal supports such as family, friends, community, and support groups to reduce negative emotional problems. Sharing their experiences reportedly strengthened the endurance and resolve of the participants. There were several parents who felt they had grown as an individual through having a child with a disability. Those feeling better prepared for raising a child with a disability reported being more emotionally stable. The participants in this study felt their health problems were related to time, family, self-care costs, and lack of supports needed for sustaining their roles as long-term caregivers. It should be noted most the coping strategies caregivers used in this study would be considered as emotion-focused. As stated earlier, the use of emotion-focused coping strategies has shown to be less effective in reducing caregiver stress than problem-focused strategies with caregivers (Smith, 2008).

In another study, Kenny & McGilloway (2007) found similar results as Smith (2008) in respect to caregiving tasks and maladaptive behaviors increasing the stress of caregivers. This mixed-methods study investigated 32 caregivers of children less than 16 years of age diagnosed with learning disabilities living in Dublin, Ireland. The study also investigated how parents felt about their caregiving role. They compared internalized and externalized strain and found that internalized strain caused caregivers greater strain in their lives than externalized strain. They did not compare gender differences in strain due to not having sufficient male participants but did find a marginal, yet significant relationship between levels of strain in those under 45 and over 45 years of age (p<.05). There was no significant difference between the strains of those caring for children with and without physical disabilities.

Kenny & McGilloway (2007) also found a significant relationship between maladaptive behaviors and caregiver stress in their mixed-methods study. Although ‘social care’ tasks showed moderately more negative affects on caregiver strain than managing problem behaviors, both showed a significantly positive correlation with caregiver strain (p<.01). Just over one third (36%) stated that they were happy or satisfied with the assistance provided them by their social worker. Of the caregivers that sought nursing or psychological assistance, only a few (38% and 18%, respectively) were satisfied with the support. This study found low satisfaction with formal supports provided in both the amount provided as well as quality. Informal supports such as spousal support (97%), other children (94%), other parents (74%), respite care (73%), and unstated 'other informal supports' rated higher than 50% approval rating. Only one informal support had less than a 50% rating, care support group (20%). More than two-thirds of the participants (*n*=21) sought professional support but only 14% (*n*=3) of those were satisfied with the services. Less than half of those who sought professional assistance felt the information received in relation to the age-related changes in their children's condition was sufficient.

Kenny & McGilloway (2007) found most of the participants appeared to find activities/hobbies on their own and participated in regular family activities (81%). Sixty-nine percent of the participants did not feel their children would be able to cope with their own lives and did not feel support systems were set up to provide quality lives for their future wellbeing. When asked about their child's life after the caregiver was no longer available to assist, 63% expressed concern and 41% refused to even think about the future. When asked about their own lives, 91% were satisfied and only 4% felt their caregiving role was restricting their lives. These parents found positive outcomes in their lives as caregivers. Parents felt the experience had helped them grow as individuals and in helping to cope better. Their confidence, openness, and honesty increased while judgmental attitudes, materialism, and selfishness decreased. This study showed that although caregiving for an individual with IDD can be stressful, one can gain positive and rewarding experiences while caregiving for their children.

In a study in the United Arab Emirates, Khamis (2007) found only one child characteristic as a significant predictor of caregiver stress; age of the individual with ID. The results of this study suggested that caregivers adapted as the child aged, which helped reduce stress and psychological distress. Khamis' study included 225 parents (113 fathers, 112 mothers) on three predictors of stress: child characteristics, parents' sociodemographics, and family environment. The age range of participants (caregivers) in this study was wider than most studies, from 21 to 85 years of age. The age range of the children was from 1 to 23 years of age. The study found these three predictors accounted for over 36% of the variance for parental stress (28.4%, 4.2%, and 3.7% respectively) and over 22% of the psychiatric symptomatology variance. Regarding child characteristics, the child's age was found to be the only significant factor associated with parental stress.   Of sociodemographics, there was only one significant predictor of stress, fathers working. There was a significant negative relationship between fathers working and stress. Personal growth (Family Environment), such as independence, recreationally-active, intellectual-cultural orientation, and achievement orientation, all were significant predictors of stress. The more this group was involved in the first three activities, the less stress they experienced. There proved to be a positive relationship between caregiver stress and those seeking achievement.

In psychological distress; the three predictors accounted for 22.5% of the variance: 10.8%, 7.9%, and 2.8%, respectively. Age (negative relationship) and severity of the disability (positive relationship) were significant predictors of psychological distress of the caregiver. Family income (sociodemographic) had a negative relationship with caregiver psychological distress. None of the environmental variables had a significant relationship with caregiver psychological distress.

Murphy et al. (2007) reported caregiver distress with regards to meeting the day-to-day needs of their children with disabilities. This combined with the uncertain future of their children resulted in caregivers reporting emotional distress. The questionnaire for this qualitative study of 28 caregivers from rural, urban, and suburban communities in Utah provided examples of situations creating distress for the caregiver because of the uncertainty of 'what is going to happen next'. These issues included: being called from school to immediately come to and remove the child; and respite time being disrupted by telephone calls about problems with their children. Many reported respite care as being problematic. This was either due to 1) not having enough respite assistance to 2) respite care not being worth all the preparation time involved. Caregivers experienced stress also from the need to constantly advocate for the rights of their children. It was claimed that this often resulted from a lack of sensitivity of the public and medical field.

Poorer health in general of caregivers was reported in the Murphy study. Most of the caregivers reported one or more chronic health conditions that they claimed were directly related to the responsibilities of long-term care of the individual. They experienced both chronic fatigue as well as sleep deprivation. Mothers reported more often than fathers as being primary caregivers, as well as having most the emotional and physical problems. Health concerns were not confined to present day; they were also concerned about the future of the individual. Caregivers felt their own deteriorating health could have a negative affect on the health of the individual in the future because they would no longer be able to meet the individual's needs (Murphy et al., 2007).

McConkey et al. (2008) investigated three indicators (family functioning, mental health, and child-related stress) of 209 mothers' sense of well-being. The mixed-methods study also investigated coping strategies through professional and informal supports and those variables that have negative impact on mothers' well-being. The study involved Irish, Taiwanese, and Jordanian mothers of children diagnosed with severe intellectual disabilities ages 5-18. The intentions were to use results of the study to help nurses provide sufficient care to parents of children with intellectual disabilities.

Mothers from all three countries (Ireland, Taiwan, and Jordan) were found to have poor mental health; increased levels of stress related to their child; and poor family functioning. Results suggest that regardless of culture, caregiving for a child with intellectual disabilities impacts families in very similar ways. The researchers went on to emphasize that the belief that families from different countries are somehow buffered from the negative effects of caregiving for a child with an intellectual disability is naïve. Yet, it was suggested that although caregivers from different countries may have similar risk factors, there are within-group differences for these risk factors and that each country should have an array of services that are based on individual needs of families and not simply provide generic, ‘canned’ programs due to those individual differences (McConkey et al., 2008).

Smith et al. (2008) studied the impact of core symptoms of autism and coping strategies of mothers caring for toddlers (*n*=153) and adolescents (*n*=201) diagnosed with ASD. Mothers of both age groups showed to have significant signs of stress. One third of the mothers also qualified as having diagnoses of clinical depression. Mothers of adolescents had higher levels of anger than did those of toddlers. Mothers of adolescents also used the coping technique of behavioral disengagement more than those with toddlers. In both findings, the researchers suggested that one reason for these results was related to the same frustrations of raising any adolescent, with or without a disability. Another good reason for frustration level differences in the two groups was the size differences of the adolescent versus the toddler. It was posited that mothers are simply better able to handle behaviors of a toddler due to small size and lack of strength than mothers of the larger adolescents. This feeling of lack of control could cause the mother to use an emotion-focused coping technique rather than a problem-focused technique (Folkman, 1984).

In relation to emotion-focused and problem-focused coping variables, Smith et al. (2008) stated that positive reinterpretation (problem-focused) and behavioral disengagement (emotion-focused) were strong predictors for mothers' well-being. Positive reinterpretation was positively related to mothers' well-being while behavioral disengagement was negatively related. Two other problem-focused strategies (active coping and planning) significantly predicted personal growth but was not a significant predictor of anger or depression. Researchers also suggested that one aspect to consider was time of diagnosis of the child. Mothers appeared to have better well-being when there was an early diagnosis.

The findings of Pozo et al. (2014) had similarities to other studies concerning individual behavior and caregiver stress. Pozo studied the quality of life and psychological well-being of fathers (*n*=59) and mothers (*n*=59) of individuals diagnosed with ASD. Just fewer than 50% of the mothers and over 88% of the fathers were employed outside the home. The Double ABCX model was used in the study. Severity of the disorder, behavior problems, social support, sense of coherence (SOC), and coping strategies were the topics of interest in the study. The study found that behaviors had a negative indirect effect on the families' ability to adapt and form a sense of cohesiveness. The results of the study also showed that mothers and fathers who reported social supports sufficient in handling the day-to-day demands of the children were significantly more positive about their quality of life.

Mothers of children with more severe disorders were significantly less satisfied with their quality of life than fathers, yet fathers also showed a negative relationship between severity of the disorder and quality of life. Fathers who showed a high perception of quality of life tended to use techniques that disengaged them from the stressful situations. The researcher hypothesized that one of these techniques involved working more hours outside the home to enable the avoidance of any behaviors. Yet, for both mothers and fathers, avoidance had a negative relationship with psychological well-being.

The data showed a negative relationship between severity of the disability with the adaptability skills of the mother and a negative relationship between behavioral issues and adaptability skills of the father. Adaptation and avoidance were the coping mechanisms for psychological well-being associated with fathers, while remaining positive and focusing on the problem were the coping strategies mainly associated with mothers.

A sense of connection was positively associated with both mothers and fathers in family quality of life and psychological well-being. Those caregivers who had a strong sense of connection had a stronger sense of family quality of life as well as higher levels of psychological well-being than those without a sense of connection. Social support was positively associated only with family quality of life. With children; characteristics, behaviors and avoidance had a positive relationship. This would again suggest that techniques in addressing behaviors are vital in attempting to lessen maladaptive behaviors of individuals with IDD.

Prader-Willi Syndrome Stress Studies

Stress within the family is greatly influenced by limitations and characteristics of the child (van der Borne, 1999; Khamis, 2007). The needs and supports of individuals with PWS change as they age. There is considerable emotion during the diagnostic stage (Hogg et al., 2001; Cantwell-Bartl, 2006) creating the beginning of a build-up of stressors. These stressors may continue well into adulthood. As a child ages and displays more maladaptive behaviors, there tends to be a high positive correlation with caregiver stress. Examples of these behaviors are self-injurious behaviors and sedentary lifestyle of individuals with intellectual disabilities (Oliver et al., 2010).

The ever-changing needs and resources for caregivers of individuals with PWS would also influence stress since external factors are often catalysts to maladaptive behaviors in individuals with PWS (Woodcock et al., 2011).

There have been very few studies regarding the stress of caregivers for individuals with Prader-Willi syndrome. There are even fewer studies of this population within the United States. The previous studies involving PWS were unable to recruit the large number of participants that the PWS (USA) has potential to provide in the present research. The small sample sizes of several of these studies created problems with reliability of these studies (van den Bourne, 1999; Wulffaert et al., 2009; Thomson, 2011; Mazaheri et al., 2013). Along with difficulty in recruitment issues, most of the studies required investigating caregiver stress with various other disabilities along with PWS. Many included another genetic syndrome involving the 15th chromosome; Angelman syndrome (van den Bourne, 1999; Thomson, 2011). The present study only examined the results involving the caregivers of individuals with PWS.

Hodapp et al. (1997) studied the stress and support levels for 42 families with children diagnosed with PWS between 3 and 18 years of age. Participants were recruited by the Prader-Willi Foundation of California and the Prader-Willi Alliance (New York-New England). Of the children, 16 were male and 26 females, and all lived at home. IQ's ranged from just below average (90) to mild (55). Of the 42 participants, 39 were mothers, 3 were fathers, and 19 states were represented. Families and friends provided most of the support to these families (7.5 supporters per family) with only 8% of support coming from professional support systems. Although many were reasonably satisfied with their supports, many reported no tangible or service support.

Findings suggested that parent-family problems and pessimism produced the most stress. The factors shown to create the most familial stress were: issues of their child overeating, skin-picking, sleep disorders, and hoarding. Many caregivers were experiencing sadness and disappointment, along with concern about their family and the future of their child with PWS. Unfortunately, no discussion concerning coping skills (other than the use of social support) were discussed in this study.

Van den Borne et al. (1999) investigated caregivers of individuals diagnosed with PWS and Angelman syndrome. The aim of the study was to identify psychosocial issues of parents, along with coping strategies. This cross-sectional study was conducted in the Netherlands with mothers and fathers responding separately to the self-report questionnaire mailed to them. Members of the Dutch Prader-Willi/Angelman Parent Association were participants in the study. Of the 62 responses provided by caregivers, 34 were caregivers of individuals with PWS; 29 who still lived at home with the parents. The other five lived in institutional settings or group homes.

Not including demographic items, the questionnaire used in the study identified questions concerning: uncertainty (*n*=19); negative feelings (*n*=14); depression (*n*=10); loss of control (*n*=12); self-esteem (*n*=12); and coping strategies (*n*=23). The results suggested that parents lacked information in various aspects of their children's syndrome: consequences of the syndrome; development of the child; and role of education. Caregivers worried about the condition worsening as well as challenges facing the individual in the future, including loneliness. Parents feared losing their temper or not being taken seriously by others whenever expressing themselves. These parents reported low control of their lives, especially in the areas of handling their affairs and finances.

Depression, concern for the future, constantly being ‘hunted’, and being irritable was a concern among mothers. Caregivers possessed low levels of self-esteem while having high concern of their child’s psychosocial well-being in the future since they felt the individual would eventually be confronted more often with their own limitations and confronted more intensely. Coping strategies related to avoidance through cognitive and behavioral strategies, created higher feelings of fear for them. The author suggested this feeling was due to the caregivers’ struggle to accept the problems related to the syndrome. It is worth noting that in two-parent households, fathers implementing active problem solving techniques displayed higher levels of self-esteem than those not implementing those techniques. Mothers’ self-esteem did not differ whether they used active problem solving techniques or not. Furthermore, mothers’ sense of burden was much higher than the fathers. Scores in the areas of fear, negative consequences for their child, depression, loss of control, and uncertainty about assistance in problem solving were much higher for the mothers than the fathers.

Wulffaert (2010) compared the stress levels of mothers of children with PWS to mothers of children diagnosed (n=5) with Angelman syndrome (n=24). The children being cared for were between the ages of 2-12. Fewer percentages (26%) of the mothers of children diagnosed with PWS had high levels of stress when compared to mothers caring for children diagnosed with Angelman syndrome (58%). Unlike other studies, there was no significant association between parenting stress and behavior problems of children with PWS. Other conclusions were that there was no association between stress levels of the mothers and ages of the children, and cognitive level which is consistent with other research (Hodapp, 1997). Wulffaert found that no more than 70% of the individuals diagnosed with PWS scored high on the overeating section of the study. This was contrary to expectations and could suggest why stress levels of the mothers of individuals with PWS were not as high as in other studies.

Thomson (2011) found in her Australian study that caregivers (*n*=21) of individuals diagnosed with Prader-Willi syndrome (*n*=5) and Angelman syndrome (*n*=11) were under considerable amounts of stress over an extended amount of time. The purpose of the mixed-methods study was to assess family stress as well as the coping strategies used by these families. The Family Stress and Coping Interview (FSCI), along with demographic information were used to assess the caregivers stress and coping skills.

Data suggested that caregivers using a wide range of coping strategies still exhibited high levels of stress, anger, and frustration, suggesting the use of different coping strategies did not lessen stress levels. Family members felt the demands of caring for the individuals were excessive and they did not have time to meet their own needs as well the other family members' needs. Caregivers also faced stress from worrying about financial problems and future lack of accommodations for the individual.

Thomson (2011) felt more participants were needed to find a correlation between coping strategies and stress. It should further be stated that this study included PWS and another genetic syndrome, Angelman syndrome. There were only caregivers of five individuals diagnosed with PWS, so less than one-third of the sample size of caregivers represented the population of the present study. Although no correlations could be determined, support groups were found to be utilized by many of the caregivers who also found them very beneficial. This study suggests that further research with larger sample sizes is warranted to determine stress levels and coping strategies.

Mazaheri et al. (2013) found caregivers of individuals with PWS had high levels of stress. The study was to assess the quality of life of mothers (*n*=12) along with siblings of the individual (*n*=13). In regard to mothers, over 72% were homemakers, of which half previously left their employment due to the need to care for the child with PWS. The variables investigated for the mothers were: overall family functioning, mothers' psychological health, the mother's perceived psychosocial and behavioral function of the individual's siblings, and quality of life of siblings. The comparison group was mothers and siblings of individuals with other forms of chronic health conditions.

Results indicated the entire family system is significantly affected by the demands placed on them in the caregiving of the individual with PWS. Mothers in the study reported higher levels of stress, worrying more, difficulty with family communication, increased family conflict, and a general poor quality of life than those in the comparison group. When compared to normative data (Derogatis, 1993), the mothers' self reported symptoms of psychological distress were well above the mean. There was a significant difference in the areas of: depression, hostility, obsessive-compulsivity, and behavioral symptoms when compared to non-clinical samples. Mothers also reported they had difficulty with cognition, motivation, anger, and lacked the ability to experience pleasure in normally pleasurable activities. Mothers in the study reported similar levels in their perception of the quality of life in their children as those without a diagnosis of PWS. The parents' perception of the siblings' quality of life were comparable to those parents of children having been diagnosed with cancer. The only significant difference was the mothers of children diagnosed with cancer reported higher levels of psychosocial health than mothers of individuals with PWS. This perception could provide some insight to the mother's own high levels of distress.

Mazaheri (2013) used a multitude of mixed-methods instruments. Mothers were required to complete several questionnaires: Brief Symptom Inventory (53-items) to assess their own psychological distress; Pediatric Quality of Life Inventory-Parent Proxy (23-items) to assess the perceived quality of life of their healthy child; Pediatric Quality of Life-Family Impact Model (36-items) to assess how the child's disability affects the family unit, and a family interview (48-items). The total of 160 questions, which over one-fourth were in interview format, possibly explains why the author stated that sample size was a limitation of the study.

Previous studies investigating the mental health status of caregivers for individuals diagnosed with PWS lacked information the present study intended to identify. Two of the five studies were conducted in the 1990's (van den Borne et al., 1999; and Hodapp, 1997). van den Borne studied caregiver depression, not stress, while Hodapp (1997) only used one form of coping strategy in his study, not the 14 being investigated in the present study. The three other studies had insufficient sample sizes to come to any true inferential conclusions (Mazaheri, 2013; Thomson, 2011; and Wulffaert, 2010). Furthermore, none of the previous studies examined how the explosive maladaptive behaviors and the crisis cycle affects caregivers' stress. The present study incorporated the assistance of a large national association, PWSA(USA), to recruit enough participants to ascertain the relationships between stress and the numerous variables of the study which also includes explosive maladaptive, behaviors and the crisis cycle.

Purpose of the Study:

The purpose of this study was to achieve an understanding of the relationship between stress in caregivers of individuals diagnosed with Prader-Willi syndrome and characteristics of the syndromes, as well as the caregiver. The study also examined the relationship between caregiver stress and their perceived supports and resources/respites. The variables being investigated were hyperphagic behaviors, crisis cycle of maladaptive behaviors, self-distraction coping strategies, active coping strategies, denial coping strategies, substance use coping strategies, use of emotional support coping strategies, use of instrumental support coping strategies, behavioral disengagement coping strategies, venting coping strategies, positive reframing coping strategies, planning coping strategies, humor coping strategies, acceptance coping strategies, religion coping strategies, self-blame coping strategies, perceived social supports, and resources/respites associated with the caregiving of an individual diagnosed with PWS (IPWS).

Research Question:

> Do hyperphagia, crisis cycle of maladaptive behaviors, self-distraction coping strategies, active coping strategies, denial coping strategies, substance use coping strategies, use of emotional support coping strategies, use of instrumental support coping strategies, behavioral disengagement coping strategies, venting coping strategies, positive reframing coping strategies, planning coping strategies, humor coping strategies, acceptance coping strategies, religion coping strategies, self-blame coping strategies, perceived social support, and resources/respites significantly predict stress among caregivers of IPWS?

Alternate Hypotheses:

- Alternate Hypothesis #1: There is a statistically significant relationship between hyperphagia behaviors and stress among caregivers of IPWS.
- Alternate Hypothesis #2: There is a statistically significant relationship between the crisis cycle of maladaptive behaviors and stress among caregivers of IPWS.
- Alternate Hypothesis #3: There is a statistically significant relationship between the use of self-distraction coping strategies and stress among caregivers of IPWS.

- Alternate Hypothesis #4: There is a statistically significant relationship between the use of active coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #5: There is a statistically significant relationship between the use of denial coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #6: There is a statistically significant relationship between the use of substance use coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #7: There is a statistically significant relationship between the use of emotional support coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #8: There is a statistically significant relationship between the use of instrumental support coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #9: There is a statistically significant relationship between the use of behavioral disengagement coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #10: There is a statistically significant relationship between the use of venting coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #11: There is a statistically significant relationship between the use of positive reframing coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #12: There is a statistically significant relationship between the use of planning coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #13: There is a statistically significant relationship between the use of humor coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #14: There is a statistically significant relationship between the use of acceptance coping strategies and stress among caregivers of IPWS.

- Alternate Hypothesis #15: There is a statistically significant relationship between the use of religion coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #16: There is a statistically significant relationship between the use of self-blame coping strategies and stress among caregivers of IPWS.
- Alternate Hypothesis #17: There is a statistically significant relationship between perceived social supports and stress among caregivers of IPWS.
- Alternate Hypothesis #18: There is a statistically significant relationship between resources/respites and stress among caregivers of IPWS.

Null Hypotheses:

- Null Hypothesis #1: There is no statistically significant relationship between hyperphagia behaviors and stress among caregivers of IPWS.
- Null Hypothesis #2: There is no statistically significant relationship between the crisis cycle of maladaptive behaviors and stress among caregivers of IPWS.
- Null Hypothesis #3: There is no statistically significant relationship between the use of self-distraction coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #4: There is no statistically significant relationship between the use of active coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #5: There is no statistically significant relationship between the use of denial coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #6: There is no statistically significant relationship between the use of substance use coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #7: There is no statistically significant relationship between the use of emotional support coping strategies and stress among caregivers of IPWS.

- Null Hypothesis #8: There is no statistically significant relationship between the use of instrumental support coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #9: There is no statistically significant relationship between the use of behavioral disengagement coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #10: There is no statistically significant relationship between the use of venting coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #11: There is no statistically significant relationship between the use of positive reframing coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #12: There is no statistically significant relationship between the use of planning coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #13: There is no statistically significant relationship between the use of humor coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #14: There is no statistically significant relationship between the use of acceptance coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #15: There is no statistically significant relationship between the use of religion coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #16: There is no statistically significant relationship between the use of self-blame coping strategies and stress among caregivers of IPWS.
- Null Hypothesis #17: There is no statistically significant relationship between perceived social supports and stress among caregivers of IPWS.
- Null Hypothesis #18: There is no statistically significant relationship between resources/respites and the stress among caregivers of IPWS.

CHAPTER 3

METHODOLOGY

Study Design

This research study used a quantitative approach and a cross-sectional design in determining how hyperphagic behaviors, crisis cycle of maladaptive behaviors, self-distraction coping strategies, active coping strategies, denial coping strategies, substance use coping strategies, use of emotional support coping strategies, use of instrumental support coping strategies, behavioral disengagement coping strategies, venting coping strategies, positive reframing coping strategies, planning coping strategies, humor coping strategies, acceptance coping strategies, religion coping strategies, self-blame coping strategies, perceived social supports, and resources/respites affected caregiver stress. The study sample was ascertained from individuals responding to various solicitations from the Prader-Willi Syndrome Association: United States [(PWSA (USA)]. PWSA(USA) members were notified by email at the beginning of the study with a notification placed in their bi-monthly newsletter, Facebook, and website after the study began. A reminder email was sent to each member approximately one month after the initial email. The survey was posted on the PWSA (USA) website for the duration of the study as well as on their Facebook page. The study data was collected from participants electronically through the administration of an online questionnaire. The description of participants, recruitment, instrumentation, data collection procedures, data analysis, and timeframe are provided in the following sections.

Participants

A convenient sample of caregivers for individuals diagnosed with PWS was selected for this study. Snowball sampling was also used after the study began. Prospective participants provided implied consent by connecting to the provided link. They, or the individual they care for, were also required to meet all of four inclusion criteria: 1) the caregiver had to be at least 18 years of age, 2) the individual they care for was required to be at least four years of age and diagnosed with PWS, 3), the individual with PWS could not live in a group home or another form of supervised living environment, and 4) the caregiver was not allowed to be paid a salary for providing care for the individual with PWS. Concerning the fourth criteria: Caregivers whose child receives financial benefits from the government (i.e. Social Security) were not considered as being paid. Primary, or secondary caregivers (i.e. parent, step-parent, grandparent, sibling, or surrogate) for an individual diagnosed with PWS could participate in the study. Nonmembers of the PWSA (USA) could participate if they met the criteria listed above but would not receive correspondence sent directly to them. Members and nonmembers were able to visit the website and follow instructions for participating in this research. Multiple caregivers of an individual could participate in the study but were required to complete separate questionnaires.

Recruitment

The Prader-Willi Syndrome Association: United States [(PWSA (USA)] is an advocate organization for individuals with Prader-Willi syndrome and their families. PWSA (USA) provides a website for individuals interested in this syndrome. There is an estimated 7,500 members of this organization of which approximately half would meet the inclusion criteria for this study. Through emails and telephone conversations, the organization agreed to be involved with the present study (See Appendix 1). The organization asked that the following information be emailed to them prior to beginning the study: Institutional Review Board (IRB) approval; purpose of study, copies of questionnaires, and a copy of the prospective participants' invitation letter. PWSA (USA) offered their assistance with structuring the final questionnaire by use of a pilot study. At the end of the final study, the PWSA (USA) also requested any formal reports (i.e. journal articles) be sent to their organization. Each participant who finished the survey was offered the opportunity to participate in a drawing for one of eight $25 Amazon gift certificates at the end of the survey. They were led to another link to enter their contact information to provide anonymity to their identity.

Pilot Study

IRB and PWSA (USA) approval for the pilot study was required prior to the beginning of the pilot study beginning. After approval was received, the PWSA (USA) emailed the online survey to PWSA(USA) members of one state within the USA. The purpose of the pilot study was to establish reliability for one set of questions that was created specifically for this study (Crisis Cycle of Maladaptive Behaviors), overall quality and clarity of the survey instrument, establish total time for completion of the survey, and to establish the data coding procedures in SPSS (Gautam, 2012). At the end of each section, pilot study participants were provided opportunities to provide their remarks and recommendations about the survey questions.

Validity was sought for the 'crisis cycle of maladaptive behaviors' portion of this study through PWSA(USA) associates. Hard copies of the 10-item questionnaire were sent to various staff members selected by the PWSA(USA) administration and support staff for content validation. The email address and telephone number of the researcher were provided to those contacted for validation purposes. The primary study began shortly after all responses to the hard-copy questionnaires, as well as the online questionnaires were completed and all recommendations by participants were addressed.

Instrumentation

Previous surveys assessing the relationship between caregiver stress and individuals with various forms of IDD and PWS have consisted of approximately 100 questions (van den Bourne, 1999; Wulffaert et al., 2010; Thomson, 2011; Mazaheri et al., 2013). The present study asked participants to answer 128 questions to assess caregiver stress. Measurement tools for the present study consisted of nine sections: demographics of caregiver and individual diagnosed with PWS; care needs of the individuals diagnosed with PWS; perceived caregiver health status; health literacy of the caregiver, hyperphagia concerns; crisis cycle of maladaptive behaviors; self-distraction coping strategies; active coping strategies; denial coping strategies; substance use coping strategies; use of emotional support coping strategies; use of instrumental support coping strategies; behavioral disengagement coping strategies; venting coping strategies; positive reframing coping strategies; planning coping strategies; humor coping strategies; acceptance coping strategies; religion coping strategies; self-blame coping strategies of the caregiver; perceived social support; and mental health status of the caregiver. The online survey platform, *Qualtrics* was used for data collection. Items included multiple choice, fill-in-the-blanks, and yes/no questions (Table 2).

Section #1(Demographic Questionnaire):

This section provides vital information about the individual, caregivers, and the environment. The eight questions about the individual diagnosed with PWS include: age; type of primary residence (2 items); gender; intellectual disability diagnosis (if any); height; weight; and if the residence provides individual access to food. There are ten questions about the caregivers and or household; age; if they are paid for caring for the individual; relationship to the individual; marital status; ethnicity; level of education; amount of care provided for the individual; annual income; number of other children living in the residence (whether biological siblings or not); and number of siblings living outside the home. The first four questions are inclusion criteria questions and if answered incorrectly the participant was sent to the end of the survey. Those questions (and correct answers) are: caregiver age (must be 18 years of age of older); is the caregiver is being paid to care for the individual (the answer must be no); age of the individual (must be at least four years old); and if the individual lives in a supervised group home or other form of supported living (the answer must be no.)

Section #2 (Individual Care Needs/Resources/Respites):

Section two has seven items about time spent between the caregiver and individual with PWS: time spent with the individual with PWS, time the individual with PWS spends at school or work; leisure time outside the home the individual with PWS participates in; three questions about respite care inside and outside of the home; and out-of-home services provided to the individual with PWS. All scoring ranges in this section are scaled from ‘0’ (no resources/respites services provided) to ‘5’ (a lot of resources/respites provided) These questions were adapted from an Australian study (Thomson, 2011).

Section #3 (Caregiver Health Status):

Section three asks four questions adapted from Lawton et al. (1982) concerning the perceived health status of the caregiver. Chronbach alpha values were calculated at .75 and .79 in a study of 251 mothers of individuals with IDD or mental health problems (Pruchno & Patrick, 1999). The questions involve overall health status, changes of status over the past three years, barriers preventing the caregiver from participating in preferred activities due to health problems, and comparing ones' own health to others of the same age. Answers to these questions range from 'very poor' 'to excellent'.

Section #4 (Health Literacy):

Section four asks three questions to assist in identifying the adequacy of caregivers in health literacy. This assists in determining if caregivers have difficulty in understanding information they read concerning PWS. The questionnaire is derived from Chew et al. (2004). The three questions asked in this questionnaire pertain to: how often other people must read the information for the caregiver; confidence levels in filling out medical forms; as well as difficulty in reading medical information. Five options are provided as answers: always, often, sometimes, occasionally, and never.

Section #5 (Hyperphagia Questionnaire):

This section has thirteen items concerning issues associated with hyperphagia (Dykens, 2007). Scoring ranges in this section are scaled to ascertain how significant are hyperphagia issues, such as '0' (little issues with hyperphagia) to '5' (significant issues with hyperphagia issues). This tool was developed to measure hyperphagia in individuals diagnosed with PWS. Through factor analyses, three significant factors accounted for 59% of the variance in hyperphagia: hyperphagia behavior (increases with age), drive (remains stable while one ages), and severity (lessens with older individuals) (Dykens et al., 2007). This was chosen instead of the often-used Food-Related Problems Questionnaire (FRPQ) because it measures strength of the hyperphagic behavior, providing a more adequate measurement of the behavior which the FRPQ does not provide (Thomson, 2011). The FRPQ is also a 16-item questionnaire verses the 13 items in this hyperphagia questionnaire. The hyperphagia questionnaire is scored on a one-to-four scale.

Section #6: (Crisis Cycle of Maladaptive Behaviors):

This section has ten questions addressing the crisis cycle of maladaptive behaviors. These include seven questions concerning the frequency, duration, and intensity of crisis cycle of maladaptive behaviors, when the IPWS first displays signs of being upset, the time it takes the behavior to become a crisis, how much time the crisis cycle takes to peak, how long does the crisis cycle stay at peak level, and the time it takes for the IPWS to completely calm down. There are also three questions in relation to how the behaviors affect the environment and how the environment is when there are no maladaptive behaviors being displayed. All scoring ranges in this section are scaled to determine severity of these behaviors and the effect on others such as ‘0’ (no issues with crisis cycle) to ‘5’ (severe issues with crisis cycle issues).

Section #7: Coping Skills (Brief COPE):

There were three variations of the COPE scale instrument considered in assessing coping strategies of the participants in this study. The original 60-item COPE instrument created by Carver, et al. (1989) has been used extensively as a flexible multidimensional coping scale. Yet, this scale was later considered time consuming so Carver created a shorter version of the COPE (Carver, 1997). The later version Carver (1997) created had fewer items (28-items) and can be used in studies that are limited by time constraints of the participants. The Brief COPE consists of 14 scales with two items per scale. All scoring ranges in this section are scaled from ‘0’ (not doing it at all) to ‘3’ (doing it a lot). The Brief COPE has efficiently been used in determining subscales analyzing adaptive and maladaptive coping strategies: Problem-focused and Emotion-focused strategies (Tuncay, 2008).

The coping strategies Hastings et al. (2005) extracted from the Brief COPE: active avoidance (emotion-focused), problem solving (problem-focused), positive coping (problem-focused), and religious/denial coping (emotion-focused) were also considered. When comparing the three options of instrumentation, it is believed the original COPE would require too much time for participants to complete the survey and Hastings' extracted version did not provide the adequate number of coping strategy variables desired for this study. The decision was made to use the Brief COPE. The 14 scales included: self-distraction coping strategies; active coping strategies; denial coping strategies; substance use coping strategies; use of emotional support coping strategies; use of instrumental support coping strategies; behavioral disengagement coping strategies; venting coping strategies; positive reframing coping strategies; planning coping strategies; humor coping strategies; acceptance coping strategies; religion coping strategies; self-blame coping strategies. Although a composite score is not part of the research question, both Problem-focused and Emotion-focused strategies were analyzed to obtain their relationship with stress.

Section #8: *Social Provisions Scale:*

This questionnaire has 24 items which assess six dimensions of perceived social supports: guidance, reliable alliance, reassurance of worth: opportunity of nurturance, attachments, and social integration (Russell & Cutrona, 1984). Each of the subscales has four items, with two being positively stated and the other two negatively worded. Lopez and Cooper (2011) claimed various research has found reliability ratings ranging from .83 to .92. This scale has been used to assess social support of numerous populations including caregivers of children. All scoring ranges in this section are scaled from '0' (Strongly disagree) to '3' (Strongly agree). Although all the individual dimensions were analyzed, only the composite score was used for answering the research question.

Section #9: Mental Health Depression, Anxiety, Stress Scale (DASS21):

This questionnaire consists of 21 self-report items concerning caregiver self-perceived mental health status in three domains: stress, depression, and anxiety. Mitchell et al. (2008) found a strong comparison of DASS-21 to clinician judgment and showed that this measure has clinical utility as a screening measure for assessing Depression, Anxiety and Stress in patients with spinal cord injuries. It was determined in that study, DASS-21 may be most useful for identifying those with GAD. Per Anastasi (1990), when assessing reliability, α must be greater than .85 to be able to make inferences while others have claimed that an α as low as .70 is sufficient (Bratas et al., 2014). For this instrument, stress and depression meet this criterion (.90 and 88, respectively) while anxiety is slightly less but still falls above the cutoff scoring of .70 stated by Bratas et al. (α=.82). The α for the total scale met criteria (.88). Another study (Asghari et al. 2008) found DASS21 to have an overall good to excellent internal consistency and good stability over time. All scoring ranges in this section are scaled from ‘0’ (Never) to ‘3’ (Almost Always).

Data Collection

Data was collected through an online Qualtrics questionnaire distributed by PWSA (USA) to their members’ email accounts. Timing of the online study was arranged to coincide with a bi-monthly PWSA(USA) newsletter which was distributed in mid-February. The newsletter was distributed in one of two different formats: by email or through the postal service, with both providing information on how to access the survey. If sent by email, the member could quickly access the hyperlink by clicking on it. If the newsletter was mailed through the postal service, the newsletter informed how to access the link on their website.

Others meeting eligibility requirements but not receiving PWSA (USA) emails or newsletters (i.e. nonmembers) could visit the website and participate in the study if they became aware of the study through other sources (snowballing). The use of online dissemination of the questionnaire was chosen for reasons such as: (1) the researcher's access to the Qualtrics software through the University of Mississippi, (2) a reduction of costs associated with printing questionnaires on paper, (3) costs of envelopes, and postage, (4) easy access to many participants, and (5) the ability for easy manipulation and exporting of data after collection.

The initial PWSA (USA) email to potential participants contained a message from the researcher briefly explaining the purpose of the study, inclusion and exclusion criteria, consent and withdrawal issues, and access to a survey link (Appendix 1). The initial email was sent to PWSA(USA) members not part of the pilot study, living in the remaining 49 states. This email was sent on November 6, 2016. A reminder email with the same information as the initial email was sent to all potential participants on January 15th. In all cases, informed consent was implied by the individual accessing the link. Each participant had the ability to enter a drawing for one of eight $25 gift certificates at the end of the study.

Data Analysis

Data were exported to a statistical software analysis program (SPSS, version 23) after all survey responses were received. The DASS21 scores were averaged (summative response scale) and Pearson Correlation Coefficient was used to determine the relationship between the independent variables. Multiple regression was used to calculate the effect of each independent variable on the dependent variable. Refer to table 2 for the summary of procedures used for data analysis.

Table 2. Summary of Data Analysis Procedures

Research Questions	Survey Item	Data Analysis Procedure
Are hyperphagia behaviors significantly related to stress among caregivers of IPWS?	Hyperphagia Questionnaire	Multiple Regression
Is the crisis cycle of maladaptive behaviors significantly related to stress among caregivers of IPWS?	Crisis Cycle of Maladaptive Behaviors	Multiple Regression
Is the use of self-distraction coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of active coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of denial coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of substance use coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of emotional support coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of instrumental support coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of behavioral disengagement coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of venting coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of positive reframing coping strategies significantly related to stress	Brief COPE	Multiple Regression

among caregivers of IPWS?

Summary of Data Analysis Procedures Continued

Research Questions	Survey Item	Data Analysis Procedure
Is the use of planning coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of humor coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of acceptance coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of religion coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Is the use of self-blame coping strategies significantly related to stress among caregivers of IPWS?	Brief COPE	Multiple Regression
Are perceived social supports significantly related to stress among caregivers of IPWS?	Social Provisions Scale	Multiple Regression
Are resources/respites significantly related to stress among caregivers of IPWS?	Individual Care Needs/Resources/Respites	Multiple Regression
Degree of Stress	DASS21	Multiple Regression

Time Frame of Study

Final IRB approval for the primary study was received in November of 2016. The initial email, reminder email, newsletter, and other dissemination of survey was begun on December 6, 2016 and ended on March 6, 2017. Data analysis was performed in March and April of 2017. Table 3 provides the timeline for the dissertation.

Table 3. Dissertation Time Line

Project Timeline	Fall 2015	Fall 2016	Spring 2017	Summer 2017
Proposal	November			
Pilot Study IRB Approval		October		
Pilot Study Data Analysis		October		
Full Study IRB Approval		November		
Final Study Data Collection Begins		December		
Final Study Data Collection Ends			March	
Study Data Analysis Begins			March	
Study Data Analysis Ends			April	
Write Final Chapters			April	
Reporting Final Results				June

CHAPTER 4

RESULTS

The purpose of this study was to determine the relationship between caregiver stress and 18 characteristics associated with PWS and their caregivers. Those variables were: 1) hyperphagia of those being cared for; 2) crisis cycle of maladaptive behaviors; 3) self-distraction coping strategies; 4) active coping strategies; 5) denial coping strategies; 6) substance use coping strategies; 7) use of emotional support coping strategies; 8) use of instrumental support coping strategies; 9) behavioral disengagement coping strategies; 10) venting coping strategies; 11) positive reframing coping strategies; 12) planning coping strategies; 13) humor coping strategies; 14) acceptance coping strategies; 15) religion coping strategies; 16) self-blame coping strategies; 17) perceived social supports; and 18) resources/respites.

Pilot Study

The IRB and PWSA (USA) approval for the pilot study was received in October, 2016. After approval was received, the PWSA (USA) emailed the online survey to PWSA(USA) members of one state in the United States. Cronbach's alpha for the crisis cycle of maladaptive behaviors sections was established (α= .82). At the end of each section, members were provided opportunities to insert their remarks or concerns about survey questions.

Twenty-six caregivers participated in the pilot study. Forty-two percent (n=11) of the participants were excluded due to not meeting the inclusion criteria. The remaining 15 participants (58%) included in the final pilot study indicated the overall quality and clarity of the survey instrument as being sufficient.

Several participants suggested being clearer with inclusion criteria and other minor changes for the primary study. The estimated time to complete the survey was changed from 25 minutes to 30 minutes due to participant recommendations in the pilot study. Any needed minor revisions were made to the primary survey instrument prior to the primary study beginning. Several comments suggested that the survey took a long time to complete but each participant stated that they understood and believed no questions needed to be excluded.

Primary Study

The purpose of this study was to examine the relationship between stress and two characteristics of individuals diagnosed with PWS: hyperphagia and maladaptive crisis cycle behaviors. Caregiver coping characteristics, perceived social supports, and resources/respites were also explored in this study.

This chapter includes: 1) socio-demographics of caregivers and those they care for; 2) resources/respites of the caregiver; 3) perceived health status of the caregivers; 4) medical literacy of caregivers; 5) hyperphagia of the IPWS; 6) behaviors creating a crisis cycle of those IPWS; coping strategies of the caregiver; and 7) perceived social supports of caregivers, 8) mental health status of caregivers.

A total of 381 individuals logged onto the survey site to participate in the survey. Among those logging in, 103 (27%) were excluded for either not finishing the survey or not meeting one of the four inclusion criteria questions. This left 278 participants (73%) who completed the survey and included in the data analysis.

Demographics of Caregivers

Most (n=164, 59%) of the participants were in the age range of 35-54 years of age. Over eighty-one percent (81.3%, n=226) were mothers. Furthermore, most (n=234, 84.2%) were Caucasian, with just under 80 percent (n=222, 89.9%) being married and many (n=78, 28.1%) with a four-year college degree. Many (n=102, 36.7%) earned more than $100,000 per year and more than a quarter of the participants (n=77, 27.7%) felt they were extremely hindered from seeking employment. Over forty-four percent (n=124, 44.6%) had no other children living in the home while over fifty percent (n=142, 51.1%) had no other children living outside the home. Over 90% (n=253, 91%) had the individual they cared for living primarily in the home with them. The demographics of those participating in the survey and those they care for are provided in Tables 4 and 5.

Table 4. Sociodemographic Characteristics of Participants (Caregivers).

Variables	n(%)
Age range	
18-25 years of age	4(1.4%)
26-34 years of age	13(4.7%)
35-54 years of age	164(59%)
55-64 years of age	70(25.2%)
65 years of age and older	27(9.7%)
Caregiver's relationship to individual	
Mothers	226(81.3%)
Fathers	33(11.9%)
Stepfathers	1(.4%)
Stepmothers	0(0%)
Grandfathers	1(.4%)
Grandmothers	5(1.8%)
Siblings	5(1.8%)
Other	7(2.5%)
Ethnicity	
African-American	9(3.2%)
Hispanic	14(5%)
Asian	3(1.1%)
Native-American160(45%)	3(1.1%)
Caucasian	234(84.2%)
Other	14(5%)

Table 4. Sociodemographic Characteristics of Participants (Caregivers), continued.

Variables	n(%)
Marital status	
Single	21(7.6%)
Married	222(79.9%)
Separated	3(1.1%)
Divorced	25(9%)
Widowed.	7(2.5%)
Education	
Less than High School	2(.7%)
High School/GED	35(12.6%)
Some College	55(19.8%)
2-year College Degree	25(9%)
4-year College Degree	78(28.1%)
Master's Degree	54(19.4%)
Doctoral Degree	15(5.4%)
Professional Degree (JD, MD)	13(4.7%)
Income	
Less than $25,000 per year	28(10.1%)
Between $25,000 and $50,000 per year	54(19.4%)
More than $50,000 and up to $75,000 per year	47(16.9%)
More than $75,000 and up to $100,000 per year	44(15.8%)
More than $100,000 per year	102(36.7%)
Other children living in the home	
None	124(44.6%)
One	77(27.7%)
Two	59(21.2%)
Three	10(3.6%)
More than three	6(2.2%)

Table 4. Sociodemographic Characteristics of Participants (Caregivers), continued.

Variables	**n(%)**
Other siblings living outside the home	
None	142(51.1%)
One	67(24.1%)
Two	38(13.7%)
Three	10(3.6%)
More than three	20(7.2%)
Prevented from working	
Not particularly at all	65(23.4%)
A little	27(9.7%)
Somewhat	48(17.3%)
Very much	61(21.9%)
Extremely	77(27.7%)
Primary residence	
Primarily lives with you	253(91%)
Primarily lives with another family member	4(1.4%)
Equally shared residence with another (ex)family member	16(5.8%)
Other	5(1.8%)

Demographics of Individual Diagnosed with PWS

From Table 5, one can find the demographics of those individuals being cared for. The average age of the individual being cared for was just under 18 ($\bar{x}$=17.56, SD=9.996) and the majority were females (n=156, 56.1%). Just under a quarter (24.5%) of the participants had an average IQ of borderline (between 70-85). The average height and weight respectively was over 4’10” ($\bar{x}$=58.62”, SD=7.63) and 147 pounds ($\bar{x}$=147.62, SD=79.29) while just under (47.5%) half the caregivers stated they always restrict the individual’s access to food.

Table 5. Sociodemographic Characteristics of Individuals Diagnosed with PWS.

Variables	**mean(+/-*sd*)**	**n(%)**
Age	17.56(9.996)	
Gender		
Male		122(43.9%)
Female		156(56.1%)
Diagnosis/IQ		
Unknown		52(18.7%)
Average range or above (IQ over 86)		40(14.4%)
Borderline (between 70 – 85)		68(24.5%)
Mild (IQ between 55 – 69)		61(21.9%)
Moderate (IQ between 40 – 54)		44(15.8%)
Severe (IQ between 25 – 39)		11(4%)
Profound (IQ of less than 25)		2(7%)
Height	58.62”(7.63)	
Weight (lbs)	147.62lbs(79.29)	
Food restricted		
Never		51(18.3%)
Rarely		19(6.8%)
Sometimes		22(7.9%)
Often		54(19.4%)
Always		132(47.5%)

Resources/Respites

Table 6 identifies responses to the items/questions for the resources/respites variable. Many (n=189, 68%) spend at least ten hours but less than 20 hours a week at home and/or transporting the individual. Just under 35% (n=97, 34.9%) of those surveyed stated that the one they care for spent over 30 hours but less than 40 hours a week out of the home at school/employment/day center without the caregiver being present. Forty-six percent (n=128) claimed the individual they care for spent no time during the week in leisure activities outside the residence without the caregiver being present (i.e. walking, team sports, shopping, bicycling, attending sporting activities and/or movies). Slightly less than 71% (n=200, 28.1%) stated that they received respite services in the past year. Of those who did receive services (n=78), (28.1%) received the respite care for more than 30 days during that year and 11.5% (n=32) received an average of care for at least one hour, but less than four hours per visit. Over 56 percent (56.5) (n=157) received no out of home services in the past year.

Table 6. Resources/Respites Provided for Caregivers

Variables	mean(*sd*)	n(%)
Average hours spent caring for the individual at home and/or transporting		
None		0(0%)
At least one but less than ten hours per week		18(6.5%)
At least ten but less than 20 hours per week		21(7.6%)
At least 20 but less than 30 hours per week		28(10.1%)
At least 30 but less than 40 hours per week		22(7.9%)
40 or more hours per week		189(68%)
Average hours the individual spent out of the home at school/employment/day center without caregiver being present		
None		41(14.7%)
At least one but less than ten hours per week		30(10.8%)
At least ten but less than 20 hours per week		25(9%)
At least 20 but less than 30 hours per week		50(18%)
At least 30 but less than 40 hours per week		97(34.9%)
40 or more hours per week		35(12.6%)
Average hours the individual being cared for spent in leisure activities outside the residence without caregiver being present		
None		128(46%)
At least one but less than ten hours per week		110(39.6%)
At least ten but less than 20 hours per week		26(9.4)
At least 20 but less than 30 hours per week		11(4%)
At least 30 but less than 40 hours per week		2(.7%)
40 or more hours per week		1(.4%)

Table 6. Resources/Respites Provided for Caregivers, continued

Variables	**mean(*sd*)**	**n(%)**
Has the individual received respite care (inside or outside the home) within the last year? *Respite care is defined as a temporary professional assistance enabling a break from caregiving of the individual.		
Yes		78(28.1%)
No		200(71.9%)
Average total days respite care was provided for the individual in the past year		
None		0(0%)
Up to one day within the past year		3(1.1%)
More than one day and up to 10 total days within the past year		19(6.8%)
More than 10 days and up to 20 total days within the past year		17(6.1%)
More than 20 days and up to 30 total days within the past year		12(4.3%)
More than 30 total days within the past year		27(9.7%)
Average hours per day receive respite care provided in the past year		
None		0(0%)
Less than one hour per day		19(6.8%)
At least one hour and up to four hours per day		32(11.5%)
More than four hours and up to eight hours per day		14(5%)
More than eight hours and up to 16 hours per day		4(1.4%)
More than 16 hours and up to 24 hours per day		9(3.2%)
Out-of-home respite services used within the past year		
None		157(56.5%)
One		53(19.1%)
Two		36(12.9%)
Three		8(2.9%)
More than three		23(8.3%)
Entire Scale	13.31(2.65)	

Self-Perceived Health Status of Caregivers

Table 7 summarizes the items/questions for the variable: Self-perceived health status of caregivers. Less than half (n=124, 44.6%) of caregivers identified their health as average while 48.9% (n=136) consider their health about the same as it was three years prior to the survey. Approximately one-third (n=91, 32.7%) believed their health did not prevent them from doing things they enjoyed. Compared to others, 35.3% (n=98) believed their own health as good.

Table 7. Self-Perceived Health Status of Caregivers of Individuals Diagnosed with PWS

Variables	**n(%)**
Perceived overall health status	
Extremely poor	7(2.5%)
Below average	36(12.9%)
Average	124(44.6%)
Below average	84(30.2)
Excellent	27(9.7%)
Health in the past three years	
Become much worse	22(7.9)
Become a little worse	97(34.9%)
Stayed about the same	136(48.9%)
Become a little better	15(5.4%)
Become much better	8(2.9%)
Health problems prevent me from doing things I want to do	
A great deal	14(5%)
Moderately	31(11.2%)
Slightly	91(32.7%)
Not at all	76(27.3%)
No health problems	66(23.7%)
Compared to others my own age, my health is	n(nn%)
Extremely poor	5(1.8%)
Poor	48(17.3%)
The same	86(30.9%)
Good	98(35.3%)
Excellent 41(14.7%)	

Medical Literacy

When asked about their ability to understand medical terms: (n=217, 78.1%) of the participants claimed "never" to need help reading medical material; (n=171, 61.5%) felt they were always confident in filling out medical forms presented to them; and 64% (n=178) never needed help in learning more about medical conditions because of difficulty understanding written information. Summaries to items/questions for participants' medical literacy is provided in Table 8.

Table 8. Medical Literacy

Variables	**n(%)**
Needing help reading medical material?	
Always	2(.7%)
Often	7(2.5%)
Sometimes	11(4%)
Occasionally	41(14.7%)
Never	217(78.1)
Confidence in filling out medical forms?	
Always	171(61.5%)
Often	77(27.7%)
Sometimes	16(5.8%)
Occasionally	7(2.5%)
Never	7(2.5%)
Problems learning more about medical conditions because of difficulty understanding written information?	
Always	7(2.5%)
Often	6(2.2%)
Sometimes	23(8.3%)
Occasionally	64(23%)
Never	178(64%)

Hyperphagia

The responses in this section suggest that hyperphagia issues with those being cared for are somewhat of an issue with the mean score being more than half of the total possible score of 70 ($\bar{x}$=37.32, SD=10.82). More than a quarter (n=76, 27.3%) of the individuals being cared for became very upset when food was denied to them, while 22.3% (n=62) bargained or manipulated for food at meals a few times a year. More than one-third of the caregivers stated that it was very hard to re-direct the individual after food was on his/her mind and surprisingly, more than half (n=150, 54%) stated that the individual never foraged/rummaged through the trash for food. More than half (58.3%, n=162) also stated that the person they cared for never got up at night to seek food, although this may be done without their knowledge.

Thirty-two percent (n=89) of the caregivers stated that the individuals are somewhat persistent in looking or asking about food after being told ‘no’ or ‘no more.’ Less than thirty percent (28.4%, n=79) of the individuals spent less than 15 minutes a day discussing food outside mealtime and 49.3% (n=137) of the individuals attempted to sneak or steal food (in various ways) a few times a week.

When others attempted to stop the individual from seeking food or discussing food, 31.7% (n=88) of them became mildly distressed, while many caregivers (n=82, 29.5%) claimed the person they cared for was extremely clever or fast in obtaining food. Almost 40% (n=39.2, n=109) of the caregivers stated that food behaviors mildly interfered with daily routines. The average age hyperphagia behaviors were first noticed was five ($\bar{x}$=5.44, SD=3.619) and 41.7% (n=116) stated that variability with the individual’s food related behaviors usually stayed about the same. Table 9 provides specific information provided by caregivers about the hyperphagia characteristics of the individuals they care for.

Table 9. Hyperphagia

Item	mean(*sd*)	n(%)
How upset the individual generally becomes when denied a desired food		
Not particularly upset at all		19(6.8%)
A little upset		66(23.7%)
Somewhat upset		73(26.3%)
Very upset		76(27.3%)
Extremely upset		44(15.8%)
How often the individual tries to bargain or manipulate to get more food at meals		
A few times a year		62(22.3%)
A few times a month		41(14.7%)
A few times a week		45(16.2%)
Several times a week		70(25.2%)
Several times a day		60(21.6%)
Difficulty in re-directing the individual away from food to other things once the individual has food on his/her mind		
Extremely easy, takes minimal effort to do so		20(7.2%)
Very easy, takes just a little effort to do so		52(18.7%)
Somewhat hard, takes some effort to do so		110(39.6%)
Very hard, takes a lot of work to do so		58(20.9%)
Extremely hard, takes sustained and hard work to do so		38(13.7%)
How the individual forage/rummage through the trash for food (that you are aware of)		
Never		150(54%)
A few times a year		62(22.3%)
1–2 times a month		21(7.6%)
1–3 times a week		26(9.4%)
4 to 7 times a week		19(6.8%)

Table 9. Hyperphagia, continued

Item	mean(*sd*)	n(%)
How often the individual gets up at night to food seek (that caregiver is aware of)		
Never		162(58.3%)
A few nights a year		41(14.7%)
1 to 2 nights a month		28(10.1%)
1 to 3 nights a week		21(7.6%)
4 to 7 nights a week		26(9.4%)
How persistent the individual in asking or looking for food after being told "no" or "no more"?		
Lets go of food ideas quickly and easily		28(10.1%)
Lets go of food ideas pretty quickly and easily		65(23.4%)
Somewhat persistent with food ideas		89(32%)
Very persistent with food ideas n(%)		63(22.7%)
Extremely persistent with food ideas		33(11.9%)
Outside of normal meal times, the individual spends talking about food or engaged in food-related behaviors		
Less than 15 minutes a day		79(28.4%)
15 to 30 minutes a day		71(25.5%)
More than 30 minutes and up to an hour a day		65(23.4%)
More than one hour and up to 3 hours a day		45(16.2%)
More than 3 hours a day		18(6.5%)
How often the individual tries to steal/sneak food or steal/sneak money/credit card to purchase food (that you are aware of)?		
A few times a year		137(49.3%)
A few times a month		50(18%)
A few times a week		32(11.5%)
Several times a week		35(12.6%)
Several times a day		24(8.6%)

Table 9. Hyperphagia, continued

Item	mean(s*d*)	n(%)
When others try to stop the individual from talking about food or engaging in food-related behaviors, it generally leads to		
No distress or upset		37(13.3%)
Mild distress or upset		88(31.7%)
Moderate distress or upset		81(29.1%)
Severe distress or upset		49(17.6%)
Extreme distress, behaviors can't usually be stopped		23(8.3%)
How clever or fast the individual is in obtaining food		
Not particularly clever or fast		42(15.1%)
A little clever or fast		45(16.2%)
Somewhat clever or fast		41(14.7%)
Very clever or fast		68(24.5%)
Extremely clever or fast		82(29.5%)
The extent food-related thoughts, talk, or behavior interfere with the individual’s normal daily routines, self-care, school, or work		
No interference		33(11.9%)
Mild interference		109(39.2%)
Moderate interference		87(31.3%)
Severe interference		40(14.4%)
Extreme interference		9(3.2%)
How old the individual was when he/she first showed an increased interest in food	5.44(3.619)	
Variability in the individual's preoccupation or interest is in food		
Hardly ever varies		40(14.4%)
Usually stays about the same		116(41.7%)
Goes up and down occasionally		93(33.5%)
Goes up and down quite a lot		18(6.5%)
Goes up and down all the time		11(4%)
Entire Scale	37.32(10.82)	

Crisis Cycle of Maladaptive Behaviors

Caregivers provided answers suggesting intensity, duration, and frequency of crisis cycle maladaptive behaviors displayed by individuals being cared for as being high in all three areas. Just over 20% (21.1%, n=59) of the caregivers claimed the person they cared for never reached a crisis stage with maladaptive behaviors but of those who do, it appears the individuals (n=135, 48.6%) become extremely upset and lose control of behavior in a nonphysical nature.

Many caregivers (n=140, 50.4%) indicated that it becomes obvious immediately when the individual is upset and it takes between one minute and five minutes for the situation to reach a crisis. Almost 25% (24.8%, n=69) stated that it takes more than a minute and up to ten minutes for the crisis to peak. More than one-third (n=102, 36.7%) stated that the individual starts showing signs of calming down between 5 and 30 minutes and 28.8% (n=80) claimed it takes between 5 and 30 minutes to completely calm down.

One concern surrounding the crisis behaviors of the individual is how it affects the environment. Many (n=117, 42.1%) of the caregivers stated the environment is somewhat pleasant prior to any crisis occurring with the individual but just under 25% (n=24.8, n=69) is extremely unpleasant during the crisis. More than one-third stated that it takes less than an hour for the environment to get back to normal after the crisis is over.

Out of a possible score of 60, the mean score for this scale was just well below half ($\bar{x}$=24, SD=8.75). This would suggest that although the crisis cycle is somewhat of a problem, it is not an extreme issue. Internal consistency for the entire scale was .836. This Cronbach alpha coefficient is sufficient under recommendations of .7 (Sharma and Petosa, 2012). Only one item would increase the reliability if taken out ‘How pleasant is the environment in your home prior to any crisis being noticed?’ The Cronbach’s alpha of the entire scale would be .897 if this item were taken out.

Table 10 lists the responses provided by the survey participants regarding intensity, duration, and frequency of crisis cycle maladaptive behaviors displayed by individuals being cared for as well as the effect on the environment before, during, and after a crisis.

Table 10. Crisis Cycle of Maladaptive Behaviors

Variable	mean(*sd*)	n(%)
How often the individual's behavior reach a crisis		
Behaviors never reach a crisis		59(21.2%)
An average of once a year		16(5.8%)
An average of once every six months		50(18%)
An average of once a month		58(20.9%)
An average of once a week		71(25.5%)
Behaviors reach a crisis on a daily basis		24(8.6%)
Usually, when the individual is frustrated he/she…		
does not become upset and stays in control of behavior		7(2.5%)
is mildly upset but stays in control of behavior.		26(9.4%)
is moderately upset but stays in control of behavior.		42(15.1%)
is extremely upset but stays in control of behavior.		26(9.4%)
is extremely upset and loses control of behavior (nonphysical)		135(48.6%)
becomes extremely upset and loses control of behavior (physical)		42(15.1%)
How long it usually takes to become obvious the individual is upset		
Not applicable (individual has never reached a crisis stage)		20(7.2%)
Immediately		140(50.4%)
Not immediate but within a minute		57(20.5%)
More than a minute and up to five minutes		36(12.9%)
More than five minutes and up to ten minutes		15(5.4%)
More than ten minutes		10(3.6%)

Table 10. Crisis Cycles of Maladaptive Behaviors, continued.

Variable	mean(*sd*)	n(%)
After noticing the individual is upset, the time it usually takes for the situation to reach a crisis stage		
Not applicable (individual has never reached a crisis stage)		51(18.3%)
Immediately		40(14.4%)
Not immediate but within a minute		62(22.3%)
More than a minute and up to five minutes		80(28.8%)
More than five minutes and up to ten minutes		30(10.8%)
More than ten minutes		15(5.4%)
After reaching a crisis stage, the time it usually takes for the situation to peak		
Not applicable (individual has never reached a crisis stage)		51(18.3%)
Immediately		54(19.4%)
Not immediate but within a minute		49(17.6%)
More than a minute and up to ten minutes		69(24.8%)
More than ten minutes and up to 30 minutes		39(14%)
More 30 ten minutes		16(5.8%)
After the situation has peaked, how long it usually takes for the individual to show signs of calming down		
Not applicable (individual has never reached a crisis stage)		44(15.8%)
Less than five minutes		23(8.3%)
Between five minutes and 30 minutes		102(36.7%)
More than 30 minutes but less than two hours		78(28.1%)
Between two hours and five hours		26(9.4%)
More than five hours		5(1.8%)

Table 10. Crisis Cycles of Maladaptive Behaviors, continued.

Variable	mean(*sd*)	n(%)
After showing signs of calming down, the time it usually takes for the individual to completely calm down		
Not applicable (individual has never reached a crisis stage)		41(14.7%)
Less than five minutes		32(11.5%)
Between five minutes and 30 minutes		80(28.8%)
More than 30 minutes but less than two hours		75(27%)
Between two hours and five hours		40(14.4%)
More than five hours		10(3.6%)
The environment in the home prior to any crisis being noticed, on the average		
Not applicable, (individual has never reached a crisis stage)		42(15.1%)
Extremely pleasant		82(29.5%)
Somewhat pleasant		117(42.1%)
A little pleasant		21(7.6%)
Not very pleasant		10(3.6%)
Not pleasant at all		6(2.2%)
On the average, how unpleasant is the environment in your home during a crisis		
Not applicable, (individual has never reached a crisis stage)		45(16.2%)
Not unpleasant at all		26(9.4%)
A little unpleasant		35(12.6%)
Very unpleasant		53(19.1%)
Extremely unpleasant		69(24.8%)
Almost intolerable		50(18%)

Table 10. Crisis Cycles of Maladaptive Behaviors, continued.

Variable	mean(*sd*)	n(%)
On the average, how long does it take for the environment to get back to normal after a crisis has ended		
Not applicable, (individual has never reached a crisis stage)		47(16.9%)
Less than an hour		104(37.4%)
Between one and five hours		86(30.9%)
More than five hours and up to ten hours		14(5%)
More than ten hours and up to 24 hours		16(5.8%)
Over 24 hours		11(4%)
Entire Scale	24(8.75)	

Coping strategies

The Brief Cope instrument used in this study has 14 coping strategies: 1) self-distraction coping strategies, 2) active coping strategies, 3) denial coping strategies, 4) substance use coping strategies, 5) use of emotional support coping strategies, 6) use of instrumental support coping strategies, 7) behavioral disengagement coping strategies, 8) venting coping strategies, 9) positive reframing coping strategies, 10) planning coping strategies, 11) humor coping strategies, 12) acceptance coping strategies, 13) religion coping strategies, 14) self-blame coping strategies. Each strategy has two questions assigned within the instrument describing how one copes with stressful evens. There are four choices of answers rating a score from 0 to 3: I haven't been doing this *at all;* I've been doing this *a little bit;* I've been doing this *a medium amount*; and I've been doing this *a lot.*

Items 1 and 19 are assigned to the first coping strategy to be discussed, self-distraction. Less than 30% of those surveyed (29.5%, n=82) stated that they have not been turning to work or other activities *at all* to take their minds off things as a coping strategy. Only 102 (36.7%) stated that they have been doing something to think about it less, just *a little bit*.

Active coping is addressed in items 2 and 7 of the Brief Cope scale: Eighty-five (30.6%) of the participants stated that they have been concentrating a *medium amount* about efforts for doing something different about the situation they are in while 36% (n=100) have been taking action *a lot* to try to make the situation better.

Denial is addressed in items 3 and 8. Only 6.5% (n=18) of those asked had been telling themselves this isn't real *a lot* while only 1.1% (n=3) have not been refusing to believe it has happened *a lot*.

Individuals may cope with their circumstances through substance use. Information about how the participants in this study may use this strategy is found in questions 4 and 11. The majority (71.6%, n=199) claimed they have not used alcohol or other drugs *at all* to make themselves feel better while a small proportion (3.2%, n=9)) stated they have not been using alcohol or other drugs *a lot* to get through the situation.

The use of emotional support is addressed in items 5 and 15. A small number (n=67, 24.1%) of the participants have not been getting emotional support from others *at all* and 11.5% (n=32) have not been getting *a lot* of comfort and understanding from someone. This would suggest that emotional support is not used often in coping with stress related to the caregiving of an individual with PWS.

Items 6 and 16 are used to assess Behavioral Disengagement. Over 62% (62.9%, n=175) of those asked stated that they have not been giving up trying to deal with the situation *at all* and 200 (72.3%) have not been giving up the attempt to cope with the situation *at all*.

Venting does not appear to be used very often by those in this study. Only 10.1% (n=28) claimed they have been saying things to let their unpleasant feelings escape *a lot* and just 28 (10.1%) stated that they have been expressing their negative feelings *a lot*.

Per results, instrumental supports have not been used much by the participants. Seventy percent (n=70) of those asked stated that they have not been getting help and advice from other people *at all* while even fewer (12.9%, n=36) claim they have been not been trying to get advice or help from others about what to do *at all*.

Positive reframing appears not to be an often-used strategy by the participants. Forty-seven (16.9%) of those asked reported not trying *a lot* to see the situation in a different light to make it seem more positive while 58(20.9%) haven't been looking *a lot* for something good in what has happened.

It does not appear that many feel they need to use the coping strategy of Self-Blame. Over one-third ((36.3%, n=101) have only been criticizing themselves *a little bit* and over one-third (43.9%, n=122) have not been blaming themselves for things that happened *at all*.

Planning is a strategy that appears to be used quite often by those caring for individuals with PWS. Of those asked, approximately one-third (34.5%, n=96) have been trying *a lot* to come up with a strategy about what to do and nearly one-third (32.7%, n=91) stated that they have been thinking hard *a lot* about what steps to take.

Humor does not appear to be a coping strategy used very much by those caring for individuals with PWS. Over 43% (n=122) of those asked claimed that they have not been making jokes about the situation *at all* and 63.7% (n=177) have not been making fun of the situation *at all*.

Many have accepted the reality of rearing an individual with PWS. Over 52% (n=146) stated they have been accepting the reality of the fact that it has happened *a lot* while 48.9% (n=136) have been learning to live with it *a lot*.

Religion appears to be used quite differently by many of the participants. Comparing those who use religion verses those who do not use religion as a coping mechanism is virtually equal, more than one third (33.8%, n=94) stated they haven't been using religion or spiritual beliefs *at all* as a coping mechanism, while 82 (29.5%) stated they did so *a lot*. Furthermore, 29.5% (n=82) have not been praying or meditating *at all* while another 29.9%(n=83) claimed they did this *a lot*.

The possible range of scores for all the coping strategies was 0-6. Table 11 displays responses provided by the participants responding to questions about how they cope while rearing individuals with PWS. The coping strategy used most was Acceptance ($\bar{x}$=4.45, SD=1.57) with Denial being used the least ($\bar{x}$=1.60, SD=1.16).

Table 11. Brief Cope/Coping Strategies

Variable	Mean(sd)	n(%)
Self-Distraction (Items 1 & 19)		
I've been turning to work or other activities to take my mind off things.		
I haven't been doing this at all		82(29.5%)
I've been doing this a little bit		77(27.7%)
I've been doing this a medium amount		59(21.2%)
I've been doing this a lot		60(21.7%)
I've been doing something to think about it less		
I haven't been doing this at all		71(25.5%)
I've been doing this a little bit		102(36.7%)
I've been doing this a medium amount		70(25.2%)
I've been doing this a lot		35(12.6%)
Entire Scale	2.60(1.81)	
Active Coping (Items 2 & 7)		
I’ve been concentrating on doing something about it.		
I haven't been doing this at all		57(20.5%)
I've been doing this a little bit		66(23.7%)
I've been doing this a medium amount		85(30.6%)
I've been doing this a lot		70(25.2%)
I’ve been taking action to make the situation better		
I haven't been doing this at all		21(7.6%)
I've been this a little bit		66(23.7%)
I've been doing this a medium amount		91(32.7%)
I've been doing this a lot		100(36%)
Entire Scale	3.58(1.78)	

Table 11. Brief Cope/Coping Strategies, continued

Variable	Mean(sd)	n(%)
Denial (Items 3 & 8)		
I've been saying to myself "this isn't real."		
I haven't been doing this at all		209(75.2%)
I've been doing this a little bit		31(11.2%)
I've been doing this a medium amount		20(7.2%)
I've been doing this a lot		18(6.5%)
I've been refusing to believe that it has happened.		
I haven't been doing this at all		248(89.2%)
I've been doing this a little bit		22(7.9%)
I've been doing this a medium amount		5(1.8%)
I've been doing this a lot		3(1.1%)
Entire Scale	1.60(1.16)	
Substance Use (Items 4 & 11)		
I've been using alcohol or other drugs to make myself feel better.		
I haven't been doing this at all		199(71.6%)
I've been doing this a little bit		45(16.2%)
I've been doing this a medium amount		25(9%)
I've been doing this a lot		9(3.2%)
I've been using alcohol or other drugs to help me get through it.		
I haven't been doing this at all		205(73.7%)
I've been doing this a little bit		44(15.8%)
I've been doing this a medium amount		20(7.2%)
I've been doing this a lot		9(3.2%)
Entire Scale	1.84(1.53)	

Table 11. Brief Cope/Coping Strategies, continued

Variable	**Mean(sd)**	**n(%)**
Emotional Support (Items 5 & 15)		
I've been getting emotional support from others.		
I haven't been doing this at all		67(24.1%)
I've been doing this a little bit		114(41%)
I've been doing this a medium amount		62(22.3%)
I've been doing this a lot		35(12.6%)
I've been getting comfort and understanding from someone.		
I haven't been doing this at all		74(26.6%)
I've been doing this a little bit		120(43.2%)
I've been doing this a medium amount		52(18.7%)
I've been doing this a lot		32(11.5%)
Entire Scale	2.38(1.75)	
Behavioral Disengagement (Items 6 & 16)		
I've been giving up trying to deal with it.		
I haven't been doing this at all		175(62.9%)
I've been doing this a little bit		72(25.9%)
I've been doing this a medium amount		18(6.5%)
I've been doing this a lot		13(4.7%)
I've been giving up the attempt to cope.		
I haven't been doing this at all		201(72.3%)
I've been doing this a little bit		58(20.9%)
I've been doing this a medium amount		14(5%)
I've been doing this a lot		5(1.8%)
Entire Scale	1.89(1.32)	

Table 11. Brief Cope/Coping Strategies, continued

Variable	Mean(sd)	n(%)
<u>Venting (Items 9 & 21)</u>		
I've been saying things to let my unpleasant feelings escape.		
I haven't been doing this at all		98(35.3%)
I've been doing this a little bit		110(39.6%)
I've been doing this a medium amount		42(15.1%)
I've been doing this a lot		28(10.1%)
I've been expressing my negative feelings.		
I haven't been doing this at all		70(25.2%)
I've been doing this a little bit		110(39.6%)
I've been doing this a medium amount		70(25.2%)
I've been doing this a lot		28(10.1%)
Entire Scale	2.20(1.68)	
<u>Instrumental Support (Items 10 & 23)</u>		
I’ve been getting help and advice from other people.		
I haven't been doing this at all		70(25.2%)
I've been doing this a little bit		117(42.1%)
I've been doing this a medium amount		46(16.5%)
I've been doing this a lot		45(16.2%)
I’ve been trying to get advice or help from other people about what to do.		
I haven't been doing this at all		74(26.6%)
I've been doing this a little bit		108(38.8%)
I've been doing this a medium amount		60(21.6%)
I've been doing this a lot		36(12.9%)
Entire Scale	2.45(1.85)	

Table 11. Brief Cope/Coping Strategies, continued

Variable	Mean(sd)	n(%)
<u>Positive Reframing (Items 12 & 17)</u>		
I've been trying to see it in a different light, to make it seem more positive.		
I haven't been doing this at all		47(16.9%)
I've been doing this a little bit		89(32%)
I've been doing this a medium amount		95(34.2%)
I've been doing this a lot		47(16.9%)
I've been looking for something good in what is happening.		
I haven't been doing this at all		58(20.9%)
I've been doing this a little bit		96(34.5%)
I've been doing this a medium amount		79(28.4%)
I've been doing this a lot		45(16.2%)
Entire Scale	2.91(1.74)	
<u>Self-Blame (Items 13 & 26)</u>		
I’ve been criticizing myself.		
I haven't been doing this at all		80(28.8%)
I've been doing this a little bit		101(36.3%)
I've been doing this a medium amount		52(18.7%)
I've been doing this a lot		45(16.2%)
I’ve been blaming myself for things that happened.		
I haven't been doing this at all		122(43.9%)
I've been doing this a little bit		93(33.5%)
I've been doing this a medium amount		33(11.9%)
I've been doing this a lot		30(10.8%)
Entire Scale	2.12(1.88)	

Table 11. Brief Cope/Coping Strategies, continued

Variable	**Mean(sd)**	**n(%)**
Planning (Items 14 & 25)		
I've been trying to come up with a strategy about what to do.		
I haven't been doing this at all		21(7.6%)
I've been doing this a little bit		65(23.4%)
I've been doing this a medium amount		96(34.5%)
I've been doing this a lot		96(34.5%)
I've been thinking hard about what steps to take.		
I haven't been doing this at all		32(11.5%)
I've been doing this a little bit		65(23.4%)
I've been doing this a medium amount		90(32.4%)
I've been doing this a lot		91(32.7%)
Entire Scale	3.82(1.73)	
Humor (Items 18 & 28)		
I've been making jokes about it.		
I haven't been doing this at all		122(43.9%)
I've been doing this a little bit		92(33.1%)
I've been doing this a medium amount		36(12.9%)
I've been doing this a lot		28(10.1%)
I've been making fun of the situation.		
I haven't been doing this at all		177(63.7%)
I've been doing this a little bit		69(24.8%)
I've been doing this a medium amount		19(6.8%)
I've been doing this a lot		13(4.7%)
Entire Scale	1.42(1.64)	

Table 11. Brief Cope/Coping Strategies, continued

Variable	Mean(sd)	n(%)
<u>Acceptance (Items 20 & 24)</u>		
I've been accepting the reality of the fact that it has happened.		
I haven't been doing this at all		21(7.6%)
I've been doing this a little bit		38(13.7%)
I've been doing this a medium amount		73(26.3%)
I've been doing this a lot		146(52.5%)
I've been learning to live with it.		
I haven't been doing this at all		14(5%)
I've been doing this a little bit		48(17.3%)
I've been doing this a medium amount		80(28.8%)
I've been doing this a lot		136(48.9%)
Entire Scale	4.45(1.57)	
<u>Religion (Items 22 & 27)</u>		
I've been trying to find comfort in my religion or spiritual beliefs.		
I haven't been doing this at all		94(33.8%)
I've been doing this a little bit		66(23.7%)
I've been doing this a medium amount		36(12.9%)
I've been doing this a lot		82(29.5%)
I've been praying or meditating.		
I haven't been doing this at all		82(29.5%)
I've been doing this a little bit		67(24.1%)
I've been doing this a medium amount		46(16.5%)
I've been doing this a lot		83(29.9%)
Entire Scale	2.85(2.34)	

Table 12 shows reliability for each coping strategy within the Brief Cope Scale, as well as the entire scale. Internal consistency for the entire scale was .773. This Cronbach alpha coefficient is considered reliable (Sharma & Petosa, 2012). The coping strategy with the highest internal consistency (substance use) had a Cronbach alpha of (α = .975) which is considered as a very high reliability level (Sharma & Petosa, 2012). The coping strategy with the lowest internal consistency score was the denial coping strategy (α=.491). A Cronbach alpha of this level is not considered as adequate per (Kuijpers et al., 2013).

Table 12. Cronbach's alpha coefficients of Brief Cope variables and entire scale

Variables	**Cronbach's alpha coefficient**
Self-distraction	.493
Active coping	.698
Denial	.491
Substance Use	.975
Emotional Support	.820
Instrumental Support	.851
Behavioral Disengagement	.729
Venting	.738
Positive Reframing	.733
Planning	.739
Humor	.791
Acceptance	.596
Religion	.919
Self-Blame	.833
Entire Brief Cope	.773

Social Supports

Weiss's Model of Social Provisions Scale (1974) has 24 items addressing the types of the supports or provisions people perceived they have at their disposal. There are six different types of social supports: Attachment, Social Integration, Reassurance of Worth, Reliable Alliance, Guidance, and Opportunity for Nurturance (Cutrona & Russell, 1987). These six provisions may be divided into two difference categories: assistance related and non-assistance related. Guidance and reliable alliance fall into the first category and relate to problem-solving. Those belonging in the non-assistance category are not directly related to problem-solving but more to self-efficacy. Bandura (1977, 1982) suggests that self-efficacy beliefs are predictive of actual coping behavior. Thus, the individuals who have supportive people bolstering their self-efficacy are expected to suffer fewer deleterious effects of stress than one whose support system does not provide such bolstering. Those functions or provisions that belong in this category are: reassurance of worth, opportunity for nurturance, attachment, and social integration.

Four questions are assigned to each form of support. Half of the items describe the presence of some form of support, while the other half describes the absence of those same forms of support. Per Weiss (1974), all six provisions are necessary for a person to feel adequately supported, although some may be more crucial during different circumstances. The aggregate of all six forms of social support/provisions will be reported in this study.

Weiss (1974) states that the construct of Attachment is one of two constructs involving emotional ties. Attachment is the emotional closeness from which one receives a sense of security and that is most often provided by the spouse of the individual. Weiss (1974) went on to claim that this provision may still be provided by other family members as well as close friends. Items 2, 11, 17, and 21 are the questions addressing the construct of Attachment. Over one third (36%, n=100) disagreed about not having close personal relationships with other people and many (n=144, 51.8%) have close relationships that provide them with a sense of emotional security and well-being. Almost half (47.5%, n=132) agreed they have close relationships that provide them with a sense of emotional security and well-being. Almost half (47.5%, n=132) felt they have a strong emotional bond with at least one other person. Over one-third (36.7%, n=102) strongly disagreed when asked if they lack a feeling of intimacy with another person. Table 13 details the answers participants provided for each of the four items for Attachment.

Table 13. Social Provisions Scale: Attachment

Variables	n(%)
I feel that I do not have close personal relationships with other people.	
Strongly disagree	88(31.7%)
Disagree	100(36%)
Agree	56(20.1%)
Strongly Agree	34(12.2%)
I have close relationships that provide me with a sense of emotional security and well-being.	
Strongly disagree	19(6.8%)
Disagree	46(16.5%)
Agree	144(51.8%)
Strongly Agree	69(24.8%)
I feel a strong emotional bond with at least one other person.	
Strongly disagree	9(3.2%)
Disagree	21(7.6%)
Agree	116(41.7%)
Strongly Agree	132(47.5%)
I lack a feeling of intimacy with another person.	
Strongly disagree	102(36.7%)
Disagree	99(35.6%)
Agree	49(17.6%)
Strongly Agree	28(10.1%)

Weiss (1974) identifies Social Integration as the other construct of the Social Provisions Scale addressing affectional ties. Social Integration is a sense of belonging to a group who shares similar interests, concerns, and recreational activities and most often are identified as friends (Weise, 1974). Items 5, 8, 14, and 22 concern the construct of Social Provisions. More than half (n=156, 56.1%) agreed they had people they believed enjoyed the same social activities they did and more than half (n=148, 53.2%) also agreed they were a part of a group of people who shared their attitudes and beliefs. Furthermore, only nine (3.2%) participants strongly agreed when asked if there was no one who shared their interests and concerns while 3(1.1%) strongly agreed there is no one who likes to do the things they do. Table 14 details participants' answers for each of the four items for Attachment.

Table 14. Social Provisions Scale: Social Integration

Variables	n(%)
There are people who enjoy the same social activities I do.	
Strongly disagree	20(7.2%)
Disagree	40(14.4%)
Agree	156(56.1%)
Strongly Agree	62(22.3%)
I feel part of a group of people who share my attitudes and beliefs.	
Strongly disagree	30(10.8%)
Disagree	43(15.5%)
Agree	148(53.2%)
Strongly Agree	57(20.5%)
There is no one who shares my interests and concerns.	
Strongly disagree	107(38.5%)
Disagree	137(49.3%)
Agree	25(9%)
Strongly Agree	9(3.2%)
There is no one who likes to do the things I do.	
Strongly disagree	93(33.5%)
Disagree	154(55.4%)
Agree	28(10.1 %)
Strongly Agree	3(1.1%)

Reassurance of Worth is a construct of the Social Provisions Scale that is non-assistance-related to problem-solving (Weiss, 1974). How one is recognized for his/her competence, skills, and value to others is the crux of this construct. This construct is highly related to self-efficacy and self-esteem (Bandura, 1977, 1982). Items 6, 9, 13, and 20 address the construct of Reassurance of Worth. Almost two-thirds (n=179, 64.4%) strongly disagreed that other people do not view them as competent and almost half (n=133, 47.8%) disagreed other people do not respect their skills and abilities. Half (n=139, 50%) of those completing the survey agreed that they have relationships where their competence and skills are recognized and 180 (64.7%) agreed there are people who admire their talents and abilities. Table 15 identifies the frequencies in which these questions were answered.

Table 15. Social Provisions Scale: Reassurance of Worth

Variables	**n(%)**
Other people do not view me as competent.	
Strongly disagree	179(64.4%)
Disagree	65(23.4%)
Agree	24(8.6%)
Strongly Agree	10(3.6%)
I do not think other people respect my skills and abilities.	
Strongly disagree	133(47.8%)
Disagree	95(34.2%)
Agree	36(12.9%)
Strongly Agree	14(5%)
I have relationships where my competence and skill are recognized.	
Strongly disagree	15(5.4%)
Disagree	35(12.6%)
Agree	139(50%)
Strongly Agree	89(32%)
There are people who admire my talents and abilities.	
Strongly disagree	12(4.3%)
Disagree	20(7.2%)
Agree	180(64.7%)
Strongly Agree	66(23.7%)

The construct of Reliable Alliances reflects how strongly people feel that someone will be there for them with tangible assistance, most often family members (Weiss, 1974). Questions 1, 10, 18, and 13 are the four questions within the scale that are related to reliable allowances. Almost half (n=128, 46%) stated they agreed they have people they can depend on to help them if they really need it and 140(50.4%) disagreed if something went wrong, no one will come to their assistance. Just over half (n=140, 50.4%) strongly disagreed there is no one they can depend on for help if they really need it while 138(49.6%) agreed there are people they can count on in an emergency. Table 16 identifies how each question was answered by participants in reliable alliances.

Table 16. Social Provisions Scale: Reliable Alliance

Variables	**n(%)**
There are people I can depend on to help me if I really need it.	
Strongly disagree	25(9%)
Disagree	30(10.8%)
Agree	128(46%)
Strongly Agree	95(34.2%)
If something went wrong, no one would come to my assistance.	
Strongly disagree	106(38.1%)
Disagree	140(50.4%)
Agree	18(6.5%)
Strongly Agree	14(5%)
There is no one I can depend on for help if I really need it.	
Strongly disagree	140(50.4%)
Disagree	106(38.1%)
Agree	22(7.9%)
Strongly Agree	10(3.6%)

There are people who I can count on in an emergency.	
Strongly disagree	15(5.4%)
Disagree	22(7.9%)
Agree	138(49.6%)
Strongly Agree	103(37.1%)

Questions 3, 12, 16, and 19 reflect how one may feel with receiving needed advice or information. This construct is labeled as Guidance. Weiss (1974) states that this support is often provided by teachers, mentors, and parents. Over one-third 109(39.2%) of those completing the survey strongly disagreed there is no one to turn to for guidance in times of stress and almost half (n=133, 47.8%) agreed there was someone they can talk to about important decisions in their life. Over 53% (53.2%, n=148) agreed there is a trustworthy person they can turn to for advice if they are having problems and 130(46.8%) disagreed there is no one they feel comfortable talking about problems with. The results of how these questions were answered are in Table 17.

Table 17. Social Provisions Scale: Guidance

Variables	**n(%)**
There is no one I can turn to for guidance in times of stress.	
Strongly disagree	109(39.2%)
Disagree	106(38.1 %)
Agree	39(14%)
Strongly Agree	24(8.6%)
There is someone I could talk to about important decisions in my life.	
Strongly disagree	16(5.8%)
Disagree	34(12.2%)
Agree	133(47.8%)
Strongly Agree	95(34.2%)
There is a trustworthy person I could turn to for advice if I were having problems.	

Strongly disagree	10(3.6%)
Disagree	22(7.9%)
Agree	148(53.2%)
Strongly Agree	98(35.3%)
There is no one I feel comfortable talking about problems with.	
Strongly disagree	109(39.2%)
Disagree	130(46.8%)
Agree	32(11.5%)
Strongly Agree	7(2.5%)

Items 4, 7, 15, and 24 address Opportunities of Nurturance a person may receive during possible stressful situations. With this construct, the need to be needed by others for wellbeing is considered very important when dealing with stressful events. This construct usually involves offspring or spouses (Weise, 1974). Cutrona and Russell (1987) suggest this is not a true social support since it involves the person aiding someone else, rather than the person receiving something from another person. Yet, opportunity for nurturance is still used in social support research because much of this research focuses broadly on how interpersonal relationships effect health and how giving and receiving help may positively enhance health through some of the same cognitive mechanisms.

Of those participants answering these items, 163(58.6%) strongly agreed there are people who depend on them for help and 192 (69.1%) strongly agreed they feel a personal responsibility for the well-being of another person. One-hundred-ninety-one (68.7%) strongly agreed when asked if there is no one who really relies on them for their well-being and 210 (75.5%) disagreed that no one needs them to care for them. Table 18 provides details about how participants responded to the questions about nurturance.

Table 18. Social Provisions Scale: Opportunity for Nurturance

Variables	n(%)
There are people who depend on me for help.	
Strongly disagree	9(3.2%)
Disagree	6(2.2%)
Agree	100(36%)
Strongly Agree	163(58.6%)
I feel personally responsible for the well-being of another person.	
Strongly disagree	6(2.2%)
Disagree	12(4.3%)
Agree	68(24.5%)
Strongly Agree	192(69.1%)
There is no one who really relies on me for their well-being.	
Strongly disagree	191(68.7%)
Disagree	76(27.3%)
Agree	4(1.4%)
Strongly Agree	7(2.5%)
No one needs me to care for them.	
Strongly disagree	210(75.5%)
Disagree	64(23%)
Agree	2(.7%)
Strongly Agree	2(.7%)

Table 19 depicts ranges, means and standard deviations for each of the six Social Provisions Scales: Attachment, Social Integration, Reassurance of Worth, Reliable Alliance, Guidance, and Opportunity for Nurturance. Each Provision had a possible range of four to 16, with all but one having observed ranges from 4 to 16. The one Provision having a different range was Nurturance, which had a range of 6 to 16. The highest mean score was received with the Nurturance provision (14.46) which also had the lowest standard deviation (1.73). The lowest mean belonged to Attachment (12.14) which also had the highest standard deviation (2.87). The mean for the entire scale was 77.13, with a standard deviation of 12.01.

Table 19. Social Provisions Scale: Observed Ranges, Means, Standard Deviations

Variables	Observed Range	Mean	SD
Attachment	4 – 16	12.14	2.87
Social Integration	4 – 16	12.21	2.52
Reassurance of Worth	4 – 16	12.90	2.35
Reliable Alliance	4 – 16	12.81	2.69
Guidance	4 – 16	12.61	2.63
Nurturance	4 – 16	14.46	1.73
Entire Scale	41 – 96	77.13	12.01

Table 20 shows reliability for each of the six subscales of the Social Provisions Scale, as well as the entire scale. Internal consistency for the entire scale was .935, which is considered highly reliable (Sharma and Petosa, 2012). The item with the highest Cronbach's alpha (Attachment) was calculated at .816. Cronbach alpha of this level is considered good according to Sharma and Petosa (2012). The social provision with the lowest Cronbach's alpha (Opportunity for Nurturance) had a score of .615, which is reliable, yet low (Kuijpers et al., 2013).

Table 20. Cronbach's alpha coefficients of Social Provisions Scales variables and entire scale

Variables	**Cronbach's alpha coefficient**
Subscales/Items	
Attachment	.816
Social Integration	.829
Reassurance of Worth	.703
Reliable Alliance	.813
Guidance	.822
Opportunity for Nurturance	.615
Entire Scale	.935

Mental Health Status (DASS-21)

The DASS-21 is a 21-item instrument which uses a 4-point Likert Scale (0= never to 3= almost always) (Abdullah et al., 2015). The essential function of this instrument is to assess the severity of the core symptoms of Stress, Depression, and Anxiety. The DASS-21 provides not only a way to measure the severity of a patient's symptoms, but a means by which a patient's response to treatment can also be measured. Because the DASS-21 is a short form of the 42-item DASS, the final score of each construct (Depression, Anxiety and Stress) needs to be multiplied by two (Gomez, 2016). Participants were asked all of 21 questions within the DASS-21 instruments.

Items 1, 6, 8, 11, 12, 14, and 18 were all questions addressing stress. Of the participants, 45.3% (n=126) found it hard to wind down sometimes, while 159(57.2%) claimed they sometimes tended to over-react to situations. A total of 120(43.2%) never found themselves using a lot of nervous energy, while 149(53.6%) found themselves getting agitated sometimes, as well as sometimes having difficulty in relaxing 139(50%). Half of the participants 139(50%) found themselves becoming intolerant with anything keeping them from getting on with what they were doing, and 156(56.1%) sometimes considered themselves as being touchy. Table 21 identifies the frequency of answers to those questions related to stress.

Table 21. Mental Health: Stress

Variables	**n(%)**
I found it hard to wind down.	
Never	32(11.5%)
Sometimes	126(45.3%)
Often	81(29.1%)
Almost Always	39(14%)
I tended to over-react to situations.	
Never	44(15.8%)
Sometimes	159(57.2%)
Often	58(20.9%)
Almost Always	17(6.1%)
I felt that I was using a lot of nervous energy.	
Never	120(43.2%)
Sometimes	108(38.8%)
Often	37(13.3%)
Almost Always	13(4.7%)
I found myself getting agitated.	
Never	42(15.1%)
Sometimes	149(53.6%)
Often	66(23.7%)
Almost Always	21(7.6%)
I found it difficult to relax.	
Never	37(13.3%)
Sometimes	139(50%)
Often	60(21.6%)
Almost Always	42(15.1%)

Table 21. Mental Health: Stress, continued

Variables	**n(%)**
I was intolerant of anything that kept me from getting on with what I was doing.	
Never	93(33.5%)
Sometimes	139(50%)
Often	43(15.5%)
Almost Always	8(2.9%)
I felt that I was rather touchy.	
Never	60(21.6%)
Sometimes	156(56.1%)
Often	48(17.3%)
Almost Always	14(5%)

Items 3, 5, 10, 13, 16, 17, and 21 of the DASS-21 are concerned with depression. Of those being asked if they experience any positive feelings at all, nearly half (133, 47.8%) stated they did, while over half 140(50.4%) stated they sometimes had difficulty working up the initiative to do things. Almost half (n=136, 48.9%) stated they never felt they had nothing to look forward to and 147(52.9%) stated they sometimes felt down-hearted and blue. Over half (n=147, 52.9%) of the participants sometimes found themselves unable to become enthusiastic and 100% (n=170) found themselves often feeling they weren't worth much as a person. Furthermore, 157(56.5%) never felt life was meaningless. Table 22 provides details for the items asked to participants about depression.

Table 22. Mental Health: Depression

Variables	**n(%)**
I could not seem to experience any positive feeling at all.	
Never	105(37.8%)
Sometimes	
Often	34(12.2%)
Almost Always	6(2.2%)
I found it difficult to work up the initiative to do things.	
Never	57(20.5%)
Sometimes	140(50.4%)
Often	55(19.8%)
Almost Always	26(9.4%)
I felt that I had nothing to look forward to.	
Never	136(48.9%)
Sometimes	95(34.2%)
Often	36(12.9%)
Almost Always	11(4%)
I felt down-hearted and blue.	
Never	64(23%)
Sometimes	147(52.9%)
Often	54(19.4%)
Almost Always	13(4.7%)
I was unable to become enthusiastic.	
Never	76(27.3%)
Sometimes	147(52.9%)
Often	37(13.3%)
Almost Always	18(6.5%)

Table 22. Mental Health: Depression, continued

Variables	n(%)
I felt I was not worth much as a person.	
Never	157(56.5%)
Sometimes	79(28.4%)
Often	28(10.1%)
Almost Always	15(5%)
I felt that life was meaningless.	
Never	195(70.1%)
Sometimes	54(19.4%)
Often	19(6.8%)
Almost Always	10(3.6%)

The third mental health construct the DASS-21 addresses is anxiety. Items number 2, 4, 7, 9, 15, 19, and 20 are questions related to anxiety. Of the participants, 118(42.4%) stated they never had dryness of the mouth and well over half 168(60.4%) expressed they never experienced breathing difficulty (e.g. excessively rapid breathing, breathlessness in the absence of physical exertion). Furthermore, 168(60.4%) claimed they never felt trembling (e.g. in the hands) and 167(60.1%) stated they never worried about situations in which they might panic and make a fool of themselves. One-hundred-sixty-three (58.6%) stated they never felt close to panic while 150(54%) believed they were aware of the action of their heart in the absence of physical exertion (e.g. sense of heart rate increase, heart missing a beat). The last question asked if the participants ever felt scared for no apparent reason. Of the 178 participants, 162(58.3%) believed they never did. Table 23 provides the responses to each item addressing anxiety.

Table 23. Mental Health: Anxiety

Variables	n(%)
I was aware of dryness of my mouth.	
Never	118(42.4%)
Sometimes	91(32.7 %)
Often	49(17.6%)
Almost Always	20(7.2%)
I experienced breathing difficulty (e.g. excessively rapid breathing, breathlessness in the absence of physical exertion).	
Never	168(60.4%)
Sometimes	81(29.1%)
Often	23(8.3%)
Almost Always	6(2.2%)
I experienced trembling (e.g. in the hands).	
Never	168(60.4%)
Sometimes	59(21.2%)
Often	15(5.4%)
Almost Always	5(1.8%)
I worried about situations in which I might panic and make a fool of myself.	
Never	167(60.1%)
Sometimes	70(25.2%)
Often	27(9.7%)
Almost Always	14(5%)
I felt I was close to panic.	
Never	163(58.6%)
Sometimes	77(27.7%)
Often	26(9.4%)
Almost Always	12(4.3%)

Table 23. Mental Health: Anxiety, continued

Variables	n(%)
I was aware of the action of my heart in the absence of physical exertion (e.g. sense of heart rate increase, heart missing a beat).	
Never	150(54%)
Sometimes	78(28.1%)
Often	43(15.5%)
Almost Always	7(2.5%)
I felt scared without any good reason.	
Never	162(58.3%)
Sometimes	80(28.8%)
Often	30(10.8%)
Almost Always	6(2.2%)

The Dass-21 Scale addresses three different mental health issues: stress, depression, and anxiety. Seven items are assigned to each mental health construct. For this study, only those specific to stress will be used. Those items are: 1) I found it hard to wind down, 2) I tended to over-react to situations, 3) I felt I was using a lot of nervous energy, 4) I found myself getting agitated, 5) I found it difficult to relax, 6) I was intolerant of anything that kept me from getting on with what I was doing, 7) and I felt I was rather touchy. This Cronbach's alpha for the stress construct (.867) was less than that for Depression (.914), but higher than the construct of anxiety (.855). The reliability for the entire scale was higher than any of the sub-constructs (.939). Table 24 summarizes Cronbach's alpha for each individual scale (stress, depression, and anxiety) and for the entire DASS-21 scale.

Table 24. Cronbach's alpha coefficients of Mental Health (DASS-21) variables and entire scale

Variables	**Cronbach alpha coefficient**
Stress	.867
Depression	.914
Anxiety	.855
Entire Scale	.939

Descriptive statistics of study variables

The resources/respites variable occupied a range from 7-21, with a mean score of 13.31(sd=2.65), which would indicate moderate resources/respites are being provided. Hyperphagia also indicated a moderate mean of 37.32 (sd=10.82) with a range from 12-68. The crisis cycle mean was 34 (sd=8.75) within a range of 5 – 39. This would fall in the mid-range of this variable. Social Provisions Scale had a range between 41 – 96 of a possible 112. The mean score for this variable was 77.13 (sd=12.01), indicating these individuals have strong social provisions.

Of the Brief Cope Scale, Acceptance received the highest mean score 4.45(sd=1.57) of coping strategies used. The coping strategy least used was Denial 1.60(sd=1.16). The Stress construct of the DASS21 has a possible range of 42 (0-42). The mean score was 15.96 (sd=8.54) which would fall within the category of mild stress (Gomez, 2015).

The stress scores suggest that 50.7% (n=141) of the participants are within the normal range of stress, the largest proportion of all levels of stress. Forty-four (15.8%) have mild stress, while 17.6% (n=49) are considered moderately stressed. The second-smallest proportion of participants are in the range of severely stressed (12.6%, n=35). Those who are considered as being under extremely severe stress were much smaller in number than any other group (3.2%, n=9). A slightly fewer number fell into the range of extremely severe stress (31.7%, n=88). This would mean that more than 66% of the participants are either in the normal range of or mildly stressed (66.5%, n=185). Descriptive statistics (range, mean, ±sd) for key variables being investigated in this study are depicted in Table 25.

Table 25. Descriptive statistics of study variables

Variables	**Range**	**Min/Max Range**	**mean(±*sd)***
Resource	15	7 – 21	13.31(2.65)
Hyperphagia	57	12 – 68	37.32(10.82)
Crisis Cycle	35	5 – 39	24.00(8.75)
Self-distraction	7	0 - 6	2.59(1.81)
Active coping	7	0 – 6	3.58(1.78)
Denial	7	0 - 6	0.59(1.16)
Substance Use	7	0 – 6	0.84(1.53)
Emotional Support	7	0 – 6	2.38(1.75)
Instrumental Support	7	0 – 6	2.45(1.85)
Behavioral Disengagement	7	0 – 6	0.89(1.32)
Venting	7	0 – 6	2.20(1.68)
Positive Reframing	7	0 – 6	2.91(1.74)
Planning	7	0 – 6	3.82(1.73)
Humor	7	0 – 6	1.42(1.64)
Acceptance	7	0 – 6	4.45(1.57)
Religion	7	0 – 6	2.85(2.34)
Self-Blame	7	0 – 6	2.12(1.88)
Social Provisions	56	41 – 96	77.13(12.01)
Stress	43	0 – 42	15.96(8.54)

Assumptions of Multiple Regression

There were four sociodemographic variables controlled for in the statistical analysis: caregiver education level, caregiver age, caregiver ethnicity, and the relationship between the caregiver and the individual being cared for. Case wise diagnostics was assessed and one outlier was removed (participant number 120).

All four assumptions were met for Multiple Regression: Independence of Errors Multicollinearity, Normality, and Linearity. The Durbin-Watson statistic tests for Independence of Observations. A result near 2.0 means the residuals are uncorrelated (Field, 2009). The results for the analysis in this study was 1.89, which is good since it is close to the score of 2.0. All variables had tolerance levels of more than 2.0, meeting an acceptable threshold for not having multicollinearity (Menard, 1995). The Variance of Inflation Factor (VIF) scores for all variables were less than ten, meeting sufficient level for not having multicollinearity (Myers, 1990). Both scatterplot and Histogram suggest Linearity and Normality of Distribution. Pearson Correlation was used to assess correlation due to the sufficient results of these four statistical analyses. Refer to table 26 for Tolerance and VIF results for all included variables and figures 1 - 3 for scatterplot, Histogram, and PP Plot.

Table 26. Tolerance and VIF (Multicollinearity)

Variables	Tolerance	VIF
Model 1		
Ethnicity	.966	1.04
Relationship	.968	1.03
Caregiver Age	.970	1.03
Annual Income	.735	1.36
Education	.747	1.34
Model 2		
Ethnicity	.902	1.11
Relationship	.865	1.16
Caregiver Age	.835	1.20
Annual Income	.600	1.67
Education	.673	1.49
Resources/Respites	.793	1.26
Hyperphagia	.564	1.77
Crisis Cycle of Maladaptive Behaviors	.578	1.73
Self-Distraction	.648	1.54
Active Coping	.425	2.36
Denial	.770	1.30
Substance Use	.774	1.29
Emotional Support	.387	2.59

Table 26. Tolerance and VIF (Multicollinearity), continued

Variables	**Tolerance**	**VIF**
Model 1		
Behavioral Engagement	.550	1.82
Model 2		
Venting	.636	1.57
Instrumental Support	.401	2.50
Positive Reframing	.568	1.76
Self-Blame	.667	1.50
Planning	.342	2.92
Humor	.740	1.35
Acceptance	.620	1.61
Religion	.717	1.39
Social Provisions Scale	.460	2.17

Figure 1. Scatterplot

Scatterplot

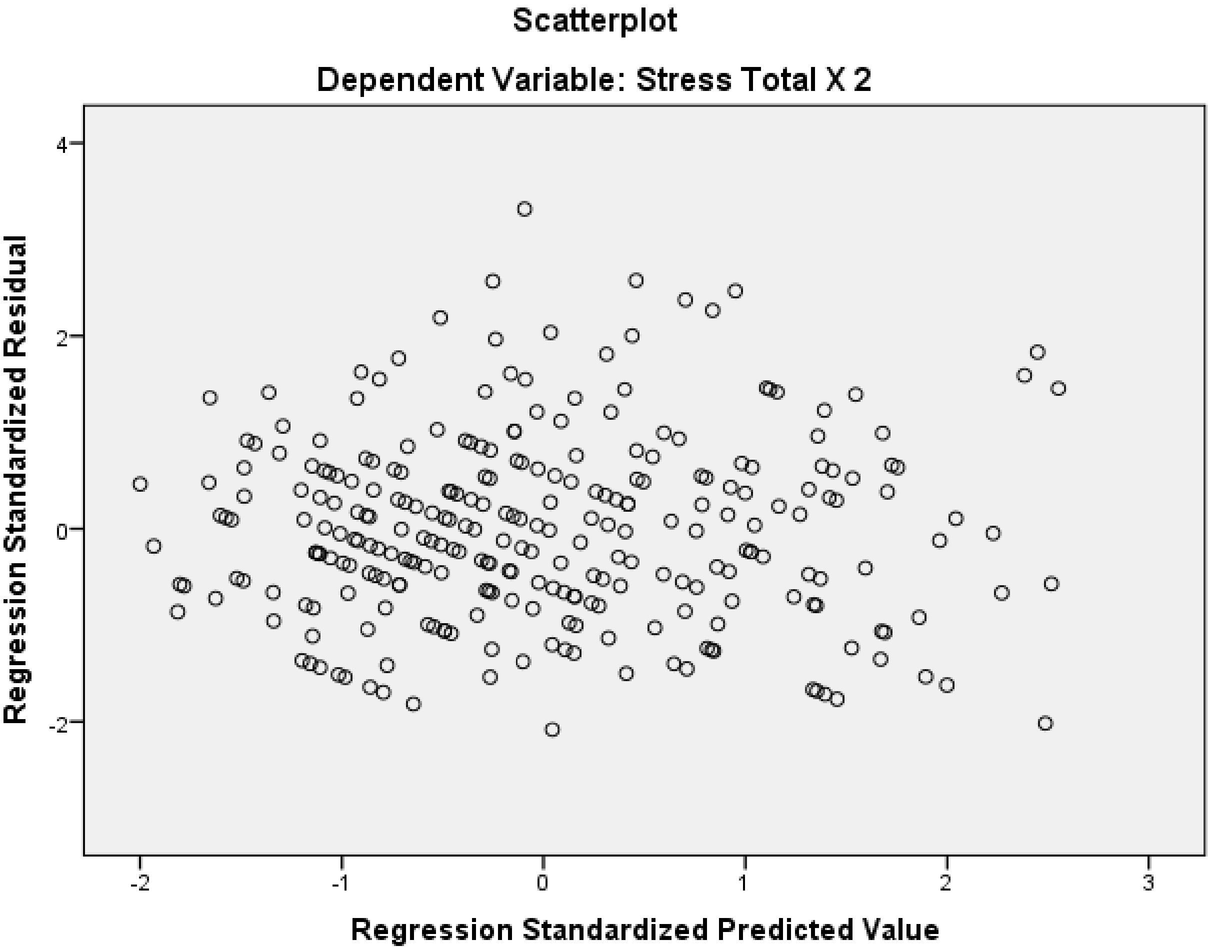


Figure 2. Histogram

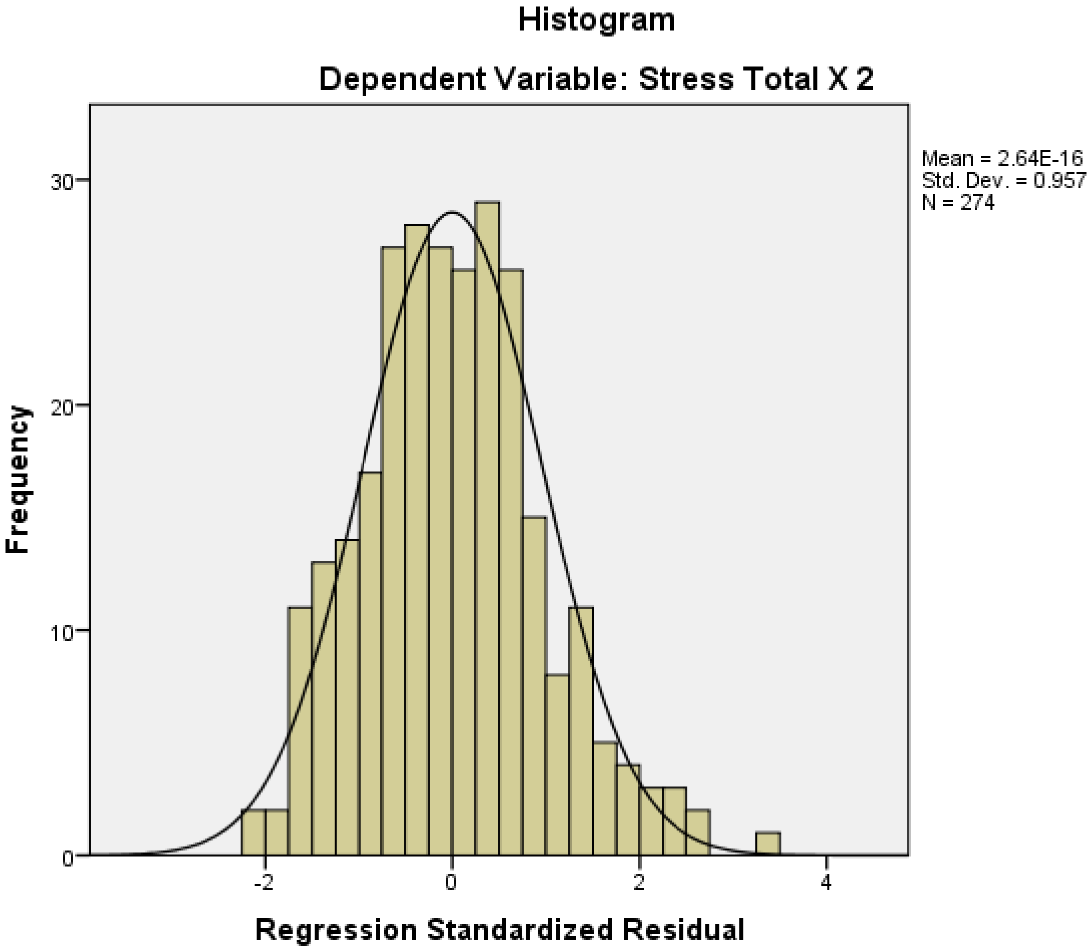
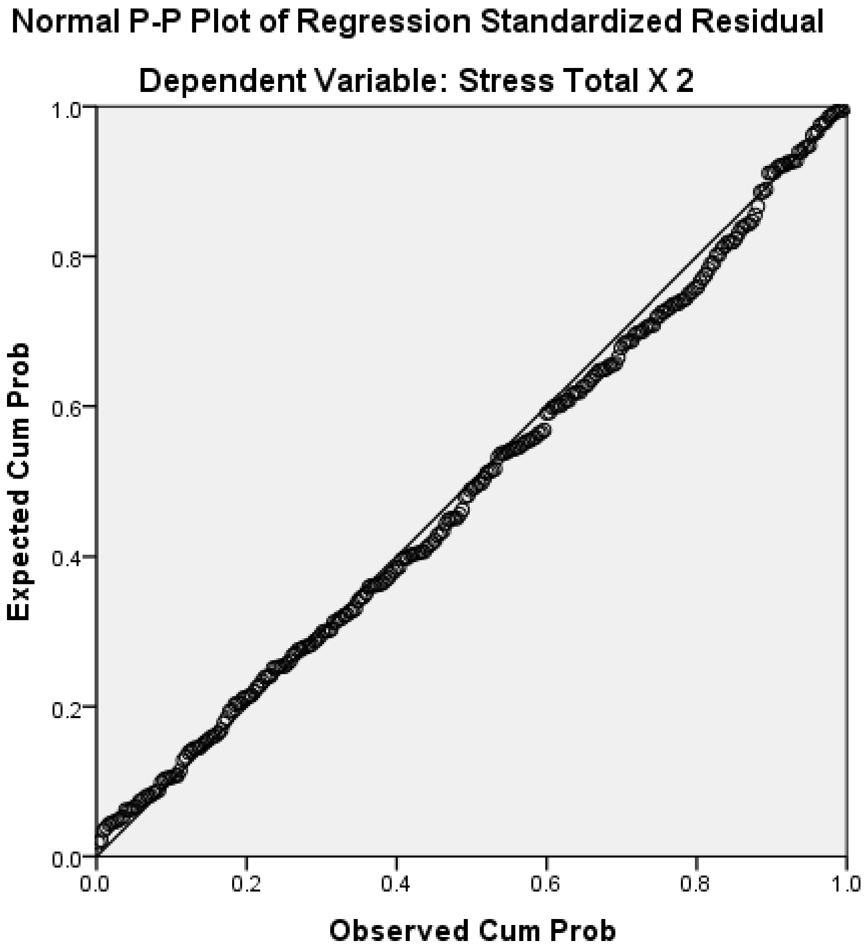


Figure 3. P-P Plot



Correlations

According to Mukaka (2012), the univariate correlations between any of the independent variables and stress did not meet the standards in meeting the threshold for moderate correlations (r = .50 - .70). In fact, only five variables in this study met the criteria for weak correlations (r= .30 - .49):

Self-Blame, Behavioral Disengagement, Venting, Social Provisions Scale, and hyperphagia. The highest correlation with stress was with one of the coping strategies (Self-Blame) which was significant (r = .487, p < .000). The variable (Acceptance) was shown to have the lowest correlation with stress (r = .030, p < .615), which was not a significant correlation. All independent variables had significant correlations, except for the following: Instrumental Support (p<.060), Emotional Support (p < .447), Positive Reframing (p < .564), Religion (p < .515), and Acceptance (p = .615). Social Provisions Scale and the Coping Strategy, Positive Reframing, were the only two variables with a negative correlation with stress. The correlations between all independent variables with caregiver stress are identified in Table 27.

Table 27. Correlations with Stress

Variable	Relationships	Pearson Correlation / Ω
Self-Blame	positive	(r = .487, p < .000)
Behavioral Disengagement	positive	(r = .442, p < .000)
Venting	positive	(r = .379, p < .000)
Social Provisions Scale	negative	(r = -.365, p < .000)
Hyperphagia	positive	(r = .332, p < .000)
Self-Distraction	positive	(r = .296, p < .000)
Crisis Cycle of Maladaptive Behaviors	positive	(r = .281, p < .000)
Denial	positive	(r = .270, p < .000)
Substance Use	positive	(r = .252, p < .000)
Resources/Respites	positive	(r = .171, p < .004)
Humor	positive	(r = .130, p < .030)
Active Coping	positive	(r = .132, p < .028)
Planning	positive	(r = .126, p < .036)
Instrumental Support	positive	(r = .113, p < .060)
Emotional Support	positive	(r = .046, p <.447)
Religion	positive	(r = .039, p < .515)
Positive Reframing	negative	(r = -035, p < .564)
Acceptance	positive	(r = .030, p < .615)

Multiple Regression

According to the Model Summary, R Square for Model 1 was equal to .069 while Model 2 equaled .421. The Model Summary also showed an R Square Change of .069 for Model 1 and R Square Change of .352 for Model 2 as both being significant (.002 and .000, respectively). The Anova Table for both Models indicated significance (.002 and .000, respectively). These results can be found in tables 28 and 29.

Table 28. Model Summary

Model	R	R Square	Adjusted R Square	Std Error of Est	R Square Change	F Change	df1	df2	Sig. F Change
1	.262	.069	.051	8.33284	.069	3.947	5	268	.002
2	.649	.421	.367	6.80399	.352	8.443	18	250	.000

Table 29. Anova

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1370.396	5	274.079	3.947	.002[b]
	Residual	18608.888	268	69.436		
	Total	19979.285	273			
2	Regression	8405.711	23	365.466	7.894	.000[c]
	Residual	11573.573	250	46.294		
	Total	19979.285	273			

Three variables showed variances of significance. The variable having the highest variance with stress was the Coping Strategy, Self-Blame (β=0.257). Social Provisions Scale was the only variable which had a significant negative score (β=-0.182). The other variable having a significant variance was Venting (β=0.183). Both Self-Blame and Venting are emotion-focused coping strategies. No problem-focused coping strategies possessed a significant variance with stress. The problem-focused coping strategy with the strongest variance was Emotional Support, possessing just over 10% of the variance (β=-.101). There were more variables with negative betas than there were correlations (n=8). The only negative beta with a significance was Social Provisions Scale. The variable with the lowest variance was a problem-focused coping strategy, Acceptance (0.002). All variances and their levels of significance are listed in table 30.

Table 30. Multiple Regression

Variables	β	Ω
Self-Blame	.257	0.000
Social Provisions Scale	-.182	0.011
Venting	.183	0.003
Self-Distraction	.113	0.060
Behavioral Disengagement	.110	0.093
Emotional Support	-.101	0.193
Caregiver Age	-.080	0.132
Hyperphagia	.078	0.224
Substance Use	.077	0.161
Instrumental Support	.076	0.321
Humor	.068	0.223
Resources/Respites	.056	0.299
Positive Reframing	.051	0.427
Religion	.050	0.380
Annual Income	-.043	0.488
Crisis Cycle of Maladaptive Behaviors	-.030	0.633
Active Coping	-.019	0.797
Planning	.019	0.818
Denial	-.016	0.775
Relationship	-.012	0.824

Table. 30. Multiple Regression, continued.

Variables	Beta Coefficients	Ω
Ethnicity	.009	0.864
Education	-.008	0.896
Acceptance	-.002	0.971

Mean Comparisons for Control Variables

Table 31 depicts the mean stress scores for the control variables of this study. Scoring for the DASS-21 is as follows: normal (0-14); mild stress (15-18); moderate stress (19-25); severe stress (26-33), and extreme severe stress (34 or greater). When comparing mean scores for the control variables, stress scores of mothers fell within the mild stress range while fathers' stress levels were in the upper normal range. Results suggest fathers' stress is slightly less than mothers'. African-Americans, Asians, and Native-Americans were in the normal range of stress, while all other groups were in the mild range of stress. Those sharing the care equally between two homes had slightly higher stress than others but still in the mild range and single parents indicated moderate stress. Analysis indicates that as the caregivers' ages increase, so do the capabilities of coping with caregiving of an individual with PWS. Those in the youngest age group (18-25 years) had the highest mean stress score (19.5) and those in the oldest age group (65 and older years) had the lowest stress (12.37). Those with an annual income of less than $25,000 were the only group with a mean stress score high enough to reach the moderate range of stress.

Table 31. Control Variables: Stress Means

Total Scores of Variable	Mean Stress Score	n/sd
Relationship with Individual		
Father	13.82 (Normal)	33/8.69
Mother	16.30 (Mild)	226/8.55
Stepfather	22.00 (Moderate)	1
Grandfather	10.00 (Normal)	1
Grandmother	11.60 (Normal)	5/6.70
Sibling	13.2 (Normal)	5/10.45
Other	20.29 (Moderate)	7/5.8
Ethnicity		
African-American	11.78 (Normal)	9/7.77
Asian	13.33 (Normal)	3/6.11
Caucasian	16.20 (Mild)	234/8.43
Hispanics	15.43 (Mild)	14/8.24
Native-American	10.00 (Normal)	3/14.00
Other	17.43 (Mild)	14/10.62
Primary Residence of Individual with PWS		
With person completing survey	15.96 (Mild)	253/8.48
With another family member	16.50 (Mild)	4/9.57
Equally shared	18.25 (Mild)	16/9.50
Other	8.4 (Normal)	5/4.56

Table 31. Control Variables: Stress Means, continued

Total Scores of Variable	Mean Stress Score	n/sd
Age of Caregiver		
18-25	19.50 (Moderate)	4/13.10
26-34	18.31 (Mild)	13/7.39
35-54	16.17 (Mild)	164/8.90
55-64	16.23 (Mild)	70/7.56
Over 65	12.37 (Normal)	27/8.08
Annual Household Income		
Less Than $25,000	20.86 (Moderate)	28/8.08
Between $25,000-$50,000	17.11 (Mild)	54/9.13
Between $50,000-$75,000	17.91 (Mild)	47/8.37
Between $75,000-$100,000	12.86 (Normal)	44/7.64
More than $100,000	14.37 (Normal)	102/8.01

Mean statistics of study variables

Those receiving the least amount of resources/respite care reported the lowest amount of stress and were in the normal range of stress while those receiving the most amount indicated the highest levels of stress (moderate). Table 32 describes the mean stress scores for the caregiver resources/respites.

Table 32. Resources/Respites: Stress Means

Total Scores of Variable	Mean Stress Score	n/sd
Resources/Respites		
7.5	13.29 (Normal)	14/7.58
9.5	12.00 (Normal)	20/6.95
11.5	15.25 (Mild)	67/7.85
13.5	16.85 (Mild)	90/8.40
15.5	14.08 (Normal)	47/8.55
17.5	20.89 (Moderate)	34/8.9
20	16.66 (Mild)	6/7.2

Those scoring in the low range with hyperphagia (12-23) scored in the normal range of stress and those scoring mid-range in hyperphagia (35-47) possessed the highest levels of stress (Extremely severe) yet those scoring in the next lowest range (24-35) had similar scores to those having the second highest hyperphagia scores (36-47). All other categories in hyperphagia were moderately stressed. Table 33 depicts the mean stress scores for the hyperphagia questions.

Table 33. Hyperphagia: Stress Means

Total Scores of Variable	Mean Stress Score	n/sd
Hyperphagia		
12-23	10.50 (Normal)	28/1.41-8.32(range)
24-35	19.22 (Moderate)	95/3.74-10.78(range)
36-47	35.52 (Extreme Severe)	74/4.83-10.49(range)
48-59	20.90 (Moderate)	45/1.41-15.28(range)
60 and over	19.33 (Moderate)	6/1.41-16.97(range)

According to analysis, stress slightly increased as crisis cycle of maladaptive behaviors increased but did not exceed the level of mild stress. Those caregivers who described the crisis cycle of the one they care for as being minor had mean stress levels in the normal range while those caring for individuals exhibiting higher levels of crisis with their maladaptive behaviors expressed a higher mean score of stress, yet only mild. Table 34 lists the mean scores for answers in the crisis cycle of maladaptive behaviors section.

Table 34. Crisis Cycle of Maladaptive Behaviors: Stress Means

Total Scores of Variable	Mean Stress Score	n/sd
Crisis Cycle of Maladaptive Behaviors		
0-10	10.11 (Normal)	38/2.83-8.64(range)
11-20	10.95 (Normal)	55/5.55-12.73(range)
21-30	14.57 (Normal)	146/4.39-11.68(range)
31-40	18.78 (Mild)	80/4.0-12.37(range)

Those using self-distraction as a coping mechanism had higher levels of stress (moderate) than those never using them (normal). All active coping strategies were within the mild range except for those never using this strategy. Those never using active coping averaged in the normal stress range. Stress increased evenly when participants used denial and substances as coping strategies. The average stress score when not using these two strategies was normal but severe stress was reported when substances were used as coping strategies a lot and extremely severe stress was reported when denial was used a lot. Emotional support stress scores were evenly dispersed throughout the participants. All mean stress scores were in the mild range. Stress increased the more behavioral disengagement was used. When behavioral disengagement was not used, stress was normal but when it was used a lot, stress almost reached the extremely severe stage. When venting was not used as a coping strategy, mean stress was normal when it was used a lot, stress average was moderate. Regardless of how much instrumental supports were used, the average stress remained in the mild range. The mean stress decreased from mild to normal as positive reframing increased. The mean stress scores increased from normal to moderate as self-blame increased as a coping mechanism, as it also did with planning. The use of humor was not widely used but of the use, the stress scores ranged from mild to moderate without regard to the answer. There was little consistency if acceptance was used as a coping strategy. Although using acceptance none-to-very little was related to mild stress, many people still used it. When used at any other level, stress remained in the mild range. Scores on religion varied little. Regardless of how the question was answered, the stress levels remained between just under mild to mild. Table 35 identifies the mean scores for all 14 coping strategies in this study.

Table 35. Coping Strategies: Stress Means

Total Scores of Variable	Mean Stress Score	n/sd
Self-Distraction (Items 1 & 19)		
0	12.38 (Normal)	48/9.07
1	12.69 (Normal)	35/8.81
2	15.25 (Mild)	56/7.53
3	17.63 (Mild)	48/8.55
4	18.39 (Mild)	42/8.0
5	17.74 (Mild)	31/6.26
6	21.00 (Moderate)	18/9.2
Active Coping (Items 2 & 7)		
0	11.16 (Normal)	19/8.85
1	16.67 (Mild)	21/11.00
2	15.28 (Mild)	36/8.49
3	15.48 (Mild)	50/8.19
4	15.86 (Mild)	57/8.0
5	18.13 (Mild)	47/7.24
6	16.58 (Mild)	48/9.09
Denial (Items 3 & 8)		
0	14.67 (Normal)	201/8.17
1	18.19 (Mild)	31/8.40
2	17.50 (Mild)	16/8.37
3	20.91 (Moderate)	22/8.25
4	20.00 (Moderate)	5/12.41

5	32.00 (Severe)	1/**
6	27.00 (Severe)	2/1.41

Table 35. Coping Strategies: Stress Means, continued

Total Scores of Variable	Mean Stress Score	n/sd
Substance Use (Items 4 & 11)		
0	14.84 (Normal)	198/8.61
1	14.75 (Normal)	8/7.55
2	17.78 (Mild)	37/6.36
3	19.14 (Moderate)	7/7.47
4	19.11 (Moderate)	18/9.03
5	28.00 (Severe)	2/.00
6	23.75 (Moderate)	8/9.47
Emotional Support (Items 5 & 15)		
0	14.35 (Normal)	51/9.62
1	17.47 (Mild)	30/9.66
2	16.12 (Mild)	86/8.77
3	16.29 (Mild)	42/7.96
4	16.13 (Mild)	32/7.07
5	14.27 (Mild)	15/6.23
6	17.36 (Mild)	22/8.03

Table 35. Coping Strategies: Stress Means, continued

Total Scores of Variable	Mean Stress Score	n/sd
Behavioral Disengagement (Items 6 & 16)		
0	12.96 (Normal)	165/7.37
1	18.72 (Mild)	36/8.00
2	20.10 (Moderate)	42/8.14
3	20.80 (Moderate)	20/8.22
4	20.40 (Moderate)	10/5.15
5	31.00 (Severe)	2/15.56
6	33.33 (Severe)	3/6.11
Venting (Items 9 & 21)		
0	10.94 (Normal)	53/7.22
1	14.00 (Normal)	47/8.31
2	16.31 (Mild)	71/6.91
3	16.92 (Mild)	50/8.50
4	20.30 (Moderate)	27/9.57
5	21.20 (Moderate)	15/8.55
6	22.00 (Moderate)	15/8.49

Table 35. Coping Strategies: Stress Means, continued

Total Scores of Variable	Mean Stress Score	n/sd
Instrumental Support (Items 10 & 23)		
0	15.64 (Mild)	44/9.90
1	14.19 (Normal)	53/8.15
2	16.32 (Mild)	69/8.65
3	13.06 (Normal)	32/7.55
4	18.86 (Mild)	37/8.74
5	18.15 (Mild)	13/5.57
6	17.33 (Mild)	30/7.62
Positive Reframing (Items 12 & 17)		
0	16.06 (Mild)	32/10.35
1	16.54 (Mild)	26/9.34
2	15.26 (Mild)	62/7.47
3	16.44 (Mild)	50/8.38
4	17.47 (Mild)	54/8.48
5	14.71 (Normal)	31/7.79
6	13.57 (Normal)	23/9.12

Table 35. Coping Strategies: Stress Means, continued

Total Scores of Variable	Mean Stress Score	n/sd
Self-Blame (Items 13 & 26)		
0	10.52 (Normal)	69/7.43
1	13.74 (Normal)	54/6.83
2	16.68 (Mild)	53/7.35
3	18.04 (Mild)	45/7.54
4	20.93 (Moderate)	15/7.52
5	22.00 (Moderate)	18/8.79
6	23.50 (Moderate)	24/8.47
Planning (Items 14 & 25)		
0	12.27 (Normal)	15/10.53
1	16.17 (Mild)	12/10.80
2	15.26 (Mild)	43/9.50
3	15.95 (Mild)	38/8.85
4	14.74 (Normal)	54/7.83
5	16.95 (Mild)	61/6.97
6	17.60 (Mild)	55/8.61

Table 35. Coping Strategies: Stress Means, continued

Total Scores of Variable	Mean Stress Score	n/sd
Humor (Items 18 & 28)		
0	14.98 (Normal)	112/8.54
1	14.75 (Normal)	61/8.13
2	16.98 (Mild)	47/9.26
3	21.04 (Moderate)	23/8.20
4	13.33 (Normal)	15/5.54
5	22.00 (Moderate)	10/7.36
6	15.80 (Mild)	10/7.39
Acceptance (Items 20 & 24)		
0	11.80 (Normal)	10/10.56
1	4.67 (Normal)	3/5.03
2	17.80 (Mild)	20/10.60
3	16.12 (Mild)	34/6.28
4	16.60 (Mild)	57/8.29
5	17.19 (Mild)	59/8.40
6	15.18 (Mild)	95/8.62

Table 35. Coping Strategies: Stress Means, continued

Total Scores of Variable	Mean Stress Score	n/sd
Religion (Items 22 & 27)		
0	14.86 (Normal)	70/8.78
1	16.69 (Mild)	29/10.22
2	17.62 (Mild)	47/8.18
3	13.18 (Normal)	17/7.21
4	15.20 (Mild)	25/5.20
5	16.55 (Mild)	22/7.44
6	16.44 (Mild)	68/9.34

The Social Provisions Scale indicated that the more support one receives, the less stress the person indicated. Those indicating the least amount of support scored in the severe range of stress. Ironically, those showing the second least amount of support had the lowest stress. Yet, in all other categories, the more support shown, the less stress was expressed by the caregiver. Table 36 describes the mean stress scores for the coping strategies.

Table 36. Social Provisions: Stress Means, continued

Total Scores of Variable	Mean Stress Score	n/sd
Social Provisions Scale		
41-50	27.2 (Severe)	10/5.26-11.31(range)
51-60	5.8 (Normal)	10/1.41-14.00(range)
61-70	18.09 (Moderate)	47/2.83-11.16(range)
71-80	17.07 (Mild)	85/5.12-11.78(range)
81-90	13.59 (Normal)	69/4.63-11.03(range)
91 and over	11.49 (Normal)	44/1.15-8.54(range)

CHAPTER 5

DISCUSSION

The primary purpose of this study was to determine the relationships between caregiver stress and 18 independent variables: resources/respites received by caregivers of individuals with Prader-Willi syndrome (PWS); hyperphagia characteristics of those being cared for; crisis cycle of maladaptive behaviors of the individuals being cared for; 14 coping strategies used by the caregivers (self-distraction, active coping, denial, substance use, use of emotional support, use of instrumental support, behavioral disengagement, venting, positive reframing, planning, humor, acceptance, religion, and self-blame), and social provisions used by the caregivers. This chapter includes: summary of findings; comparison of results with other studies; recommendations for studies in the future; limitations of the study; and a conclusion.

This was a descriptive, cross-sectional study with a total of 278 participants. Invitations to participate in the survey were sent by the national PWS organization [Prader-Willi Syndrome Association(USA)] and the data was collected via an online survey delivered through Qualtrics survey tool. The data was collected between December 2016 and March 2017. Multiple regression was used to describe the relationships between the independent variables and the dependent variable (stress).

The DASS-21 was used as the dependent variable in the present study. The DASS-21 is a tool used to describe the full range of scores on stress for a population. For example, a person scoring in the mild range only indicates that a score is above the general population norm but may be well below the level of needing to seek any help. The hierarchy of the scale is: normal, mild, moderate, severe, and extreme severe. Just over half of the participants in this study (n=141, 50.7%) scored in the normal range of stress while only 3.2% (n=9) were on the opposite end of the spectrum with extreme stress. Of those in this study, 12.6% (n=35) expressed they were severely stressed. This would leave 93 (33.5%) in the mild to moderate stress range. The mean score for stress in the present study fell just out of the normal range of stress (0-14) and within the mild range of stress of 15-to-18 ($\bar{x}$=15.96, sd=8.54). A large portion of the participants (84.1%) would be between normal to moderate stress. This would leave only 15.9% expressing some form of high stress. These results suggest that a small percentage of these participants expressed high stress compared to other studies (Hodapp et al.,1997; Mazaheri et al., 2013; Thomson, 2011; van der Borne, 1999).

Unlike this study, results of previous studies suggest that caregivers of individuals with PWS have high level of stress (Hodapp et al.,1997; Mazaheri et al., 2013; Thomson, 2011; van der Borne, 1999). Another study suggested that although caregivers of individuals with PWS had high levels of stress, they were not as high as the stress indicated by caregivers of Angelman syndrome. (Wulffaert et al., 2010). According to Wulffaert's results, 74% of the participants caring for individuals with PWS had low stress while 26% had high stress. These results are similar to the results of the present study. One difference between Wulffaert's study and the present study was that participants were asked only if they had low stress or high stress. It is unclear where those participants would be within the DASS-21. Furthermore, there were only six participants in Wulffaert's study.

Compared to the general population, Fahey (2012) states that more than half of US citizens exhibit high stress. Fahey's results would be consistent with a 2011 US survey (Stress in

America, 2012). In the 2011 study, caregivers were more likely than the general population to report high stress. On a scale from 1-10, the general population scored 5.2 on the self-reported stress scale while caregivers scored 6.5. If the results of the present study were placed on a similar scale; the percentage of those reporting high stress would be less than that in the 2011 study (20.1%).

The results of the low stress levels in this study could be a result of increased training by professionals in the field of PWS, as well as support provided by organizations such as the PWSA(USA). The PWSA(USA) provides information to individuals diagnosed with PWS and their caregivers on their website (www.pwsausa.org/) as well as their bi-monthly newsletters (The Gathered View). Numerous research has been authorized and funded through the PWSA(USA), as well. Other resources not available in the past are now available to those interested in the issues surrounding PWS. Many videos on managing behaviors and other forms of life matters surrounding PWS are now available on PWS. One on-line video specifically identifies tools caregivers of individuals with PWS can use to avoid stressful situations, such as being consistent in how they respond to the person they are caring for and the caregiver asking instead of making demands. The crisis cycle is discussed in the video, as well as discussing the need to be consistent when handling issues as they occur. Practical, as well as PWS specific examples of reinforcing good behavior are identified in this video (Roof, 2015). This video is one of many on-line videos or other types of resources these caregivers have at their disposal that were not available just a few short years ago.

Despite the results of stress levels with the participants in this study, very little could be derived from the univariate correlation analysis. None of the univariate correlations met the threshold of having a negative or positive moderate correlation of between .50 to .70 (Mukaka, 2012). In fact, most the variables were extremely weak or virtually non-existent. Of the 18 independent variables, only five met the criteria for low correlation with the dependent variable, stress

(r = .3-.5), per Mukaka (2012): Self-Blame (r = .487), Behavioral Disengagement (r = .442), Venting (r = .379), Social Provisions Scale (r= -.365). and Hyperphagia (r = .332). All these correlations were significant correlations. All but two of the 18 variables (Social Provisions Scale and Positive Reframing coping strategy) had positive correlations with stress.

Emotion-focused coping strategies have been found to be positively related to, and increase stress. van den Borne et al. (1999) found emotion-focused coping strategies related to avoidance through cognitive and behavioral strategies. Using these strategies created higher feelings of fear for them, and suggested this feeling was due to the caregivers' struggle to accept the problems related to the syndrome. Thomson (2011) suggested that caregivers in her study (n=5) used a wide variety of coping strategies, but still exhibited high levels of stress, anger, and frustration. The author suggested the use of different coping strategies did not lessen stress levels, but felt more participants were needed to truly assess correlations.

In the present study, there was virtually no use of four of the six emotion-focused coping strategies. The other two emotion-focused coping strategies were used only a little. Denial was not used much and when it was used a lot, the average stress scores advanced from normal to severe. Substance use was not used much either, but stress went from normal to moderate the more it was used. Behavioral disengagement was not used a lot and when used, average scores greatly advanced from normal to just under extreme severe. Self-Blame was the fourth coping strategy not used a lot, but when used average stress advanced from normal to moderate stress when used. Self-distraction was used moderately and ranged from normal when not used to severe when used a lot. Venting was used a moderate amount of times and stress increased from the normal range to moderate the more it was used.

All emotion-focused coping strategies produced more stress for the caregiver. Fortunately, four of these six strategies were rarely used in this study and the other two used sparingly. The fact that fewer caregivers for individuals with PWS used these maladaptive strategies would suggest that caregivers of individuals with PWS are learning the proper ways decreases stress.

Problem-focused strategies in this study were used more often than emotion-focused but only a few were used consistently. Planning and acceptance were the two problem-focused strategies that were used a lot and humor was not used much at all. Although planning was used a lot, when it was not used, the mean stress score was in the normal range and if used often, stress increased to the moderate stage. Acceptance was also used a lot but stress was also higher when used a lot than when it was used only a little. When Acceptance was used just a little bit, stress was normal but the mean stress score increased to mild the more acceptance was used. Regardless of the level of use, the average stress when using this strategy never reached higher than mild. Humor was not used a lot and the stress scores were inconsistent with its use. When not used at all, stress was in the normal range. When used a moderate amount, stress moved to the moderate level, but when its use increased to a lot, the stress levels moved back to the mild range.

All other coping strategies were used intermittently by the participants. Active coping was used moderately but the more it was used the higher stress increased, resulting in a range from normal to mild stress when it was used. Religion was used sporadically and average stress levels were normal when not used at all but remained in the mild range if used in any other capacity. Instrumental support was used moderately and average stress increased slightly the more it was used, ranging from normal to mild.

Positive reframing was used inconsistently within the participants. As the use of this coping strategy increased, stress decreased. When never used, the average stress was mild but stress decreased to normal when this coping strategy was used a lot. Emotional supports proved interesting. Although social provisions scale (social supports) indicated high levels of social support for those in this study, only 12% of the participants in this study identified emotional supports as a coping strategy they used a lot. Regardless of its use, the average stress score did not fluctuate much, with its use the stress score ranged from normal to mild.

The results of this study show that a variety of problem-focused coping strategies were used but not emotion-focused. All the emotion-focused and all but one of the problem-focused coping strategies increased average stress. The only coping strategy to decrease average stress was positive reframing. Participants using a variety of coping strategies is consistent with findings of Thomson (2011). Unlike Thomson, the average stress scores in this study averaged in the low-mild range. One may posit that there may be other ways to address the stress of raising an individual with PWS other than coping strategies. Also, many of the coping strategies were used inconsistently so the lack of consistency may have been an issue with the results.

Thomson (2011) and the present study were consistent in determining that support groups are well utilized by many of the caregivers who have found them very beneficial. Social supports had the fourth-highest correlation in this study, as well as being significant. It was also the only negative correlation other than the coping strategy: positive reframing. Although a weak correlation, the results suggest that caregivers of individuals diagnosed with PWS use their social supports in combating stress in this population.

Hyperphagia was the other variable having a positive correlation with stress, suggesting the worse the hyperphagia characteristics are, the more severe the stress for the caregiver. Although this study found hyperphagia to have a positive correlation, it was still weak. Furthermore, the beta for this independent variable (β =.078) was not significant (α=0.224). Wulffaert (2010) found similar results in his study yet this study is inconsistent with Hodapp et al. (1997).

Results of multiple regression showed the eighteen independent variables, along with sociodemographic variables accounted for 42.1% of the total variance in stress: F(23, 250)=7.894, p<0.000. The adjusted R2 provided the variance held by the eighteen independent variables, while holding constant the sociodemographic variables (R2=0.352, 35.2%). There were only three independent variables possessing significant variances with stress. Two of the variables with significant betas were emotion-focused coping strategies and possessed positive relationships: Self-Blame and Venting. All other emotion-focused coping strategies were not significant and possessed positive relationships with stress, except denial.

Unlike other studies, the present study showed several problem-focused coping strategies as having positive relationships with stress: instrumental support, humor, positive reframing, and religion. These results indicate that the more a person used these problem-focused coping strategies, the more stress they incurred. These results would be consistent with the correlation analysis. Yet, the direction of some variables was different between variables with very weak correlations and beta coefficients. This could be explained by variables being controlled for in multiple regression analysis, resulting in the positive and negative relationships between some of the variables being different between correlation analysis and multiple regression analysis.

This study is consistent with Thomson (2011) who stated that a variety of coping strategies were used by caregivers of individuals with PWS. Yet, unlike Thomson's study, caregivers in this study did not report high levels of stress. Furthermore, according to Thomson, her study had an insufficient number of participants (n=5) to infer any relationships. The present study did have enough participants to infer relationships. The results of this study would suggest the majority of the coping strategies used by the caregivers in this study did not have strong relationships with stress. The two having significant relationships were emotion-focused and used sparingly.

The other variable with a significant beta was Social Provisions Scale, or perceived social supports. This study is consistent with other studies in that it shows perceived social supports are important (Hodapp et al.,1997; van den Bourne, 1999; Thomson, 2011). These studies reported social support as beneficial in reducing caregiver stress. Thomson (2011) suggested more studies with much larger sample sizes would be beneficial in providing evidence to support these findings. This study has provided such a sample size recommended by Thomson's (2011) to support previous findings for social support. These multiple regression results would further support the correlation analysis. It would be beneficial that those caring for individuals diagnosed with Prader-Willi syndrome to make ample use of those social supports they have acquired.

This study was designed to only explore the relationships between stress and the 18 independent variables and not to compare stress of caregivers of individuals with Prader-Willie syndrome with other populations. Yet the results of this study would suggest that those caregivers in this study may not be exhibiting the same stress levels as in the previous PWS studies. The study does indicate that 15.8% of the participants do describe having severe to extreme severe stress and 33.5% of the participants experience either mild or moderate stress. This would be consistent with other studies that suggest caregivers of individuals with PWS do experience stress (Hodapp et al.,1997; van den Bourne, 1999; Wulffaert et al., 2010; Thomson, 2011; Mazaheri et al., 2013) but the degree of stress may be different. Yet, further studies comparing these individuals with other populations would prove advantageous in further explaining the relationships between stress and the various characteristics surrounding caregiving for individuals diagnosed with PWS.

The results of this study indicated no strong correlations between any of the variables and stress. Of the independent variables not related to coping, resources had the lowest correlation with stress. One previous study (Hodapp, 1997), suggested resources increase the quality of life for caregivers of individuals with PWS but the results of this study show it holds only 5.6% of the variance of stress and has a positive, non-significant correlation of .171. This could be explained by the low levels of stress of the participants and the relatively low amount of resources and respite care received by the caregivers. In the present study, it did appear caregivers had very little time away from their caregiving duties with 46% of them reporting no leisure activities the individual in their care could attend without the caregiver being present. Caregivers also suggested that caring for the individual is a full-time job, at minimum. Sixty-eight percent reported they spent forty or more hours a week transporting and caring for the individual. The most concerning issue involving resources involved lack of respite care. More than 70% of the caregivers did not receive any form of respite care over the previous year, yet the results of the study suggest stress levels were not negatively affected by this lack of resources.

Crisis cycle of maladaptive behavior possessed a strong internal consistency of .836 but held a mere beta coefficient of -3%. It also possessed a low correlation (r=.281) even though the mean stress score increased as the crisis scores increased. There were no mean stress scores that ranked higher than the mild range. This would suggest that caregivers could be using proper techniques in lessening the effects of this explosive behavior prior to the situation occurring or during the crisis.

Both the correlation (r=.332) and beta coefficient (.078) were weak for the variable Hyperphagia in this study. Through analysis, there appeared to be a relationship with stress increasing while hyperphagia characteristics increased. Those caring for individuals with minor hyperphagia characteristics, stress levels were in normal range. Those with extreme characteristics stress rose to moderate levels.

The present study is the largest to date of stress and caring for an individual with PWS. The results indicate only three items having significant relationships with stress: social provisions, self-blame, and venting. Self-blame and venting are both emotion-focused coping (maladaptive) strategies, usually resulting in increased stress. The results of this study are no different. Both had positive beta coefficients of .257 and .183, respectively. Although both had significant betas, participants did not use these coping strategies to the degree they did problem-focused strategies. This could explain why there was a negative multiple regression slope. Social provisions proved to have a significant relationship with stress with a -.182 beta. Scores on this scale suggest the individuals in the present study use social supports effectively and often. This could be the reason participants in this study have lower stress levels than studies of this population in the past.

Future research would prove beneficial in the areas of teaching strategies in using problem-focused coping strategies consistently. Results of this study suggest these caregivers are using the less efficient emotion-focused coping strategies less but may not be using the more efficient problem-focused strategies. Future research in the mediating effects of the social supports and coping strategies could enhance knowledge of the interaction these independent variables have on stress.

Limitations of Study

There are limitations to this study which need to be identified.

1) There is lack of prior research studies on the topic. Citing previous research forms a basis for literature review. Much of the previous research with stress involved different populations than caregivers of individuals with PWS. This lack of research required an exploratory approach to the topic area rather than explanatory approach. Follow-up research could build a stronger overall evidence base.
2) Self-reported data cannot be independently verified. Self-reported data has potential biases, such as selective memory and exaggeration. Both can be a result of social desirability and recall biases.
3) The incentive provided in this study also may have caused some to participate only for the chance to win a $25 gift certificate. With the incentive being the main reason for participation, the self-reported data may create a bias.
4) Although participants were recruited from a national PWS organization, only a small percentage of all caregivers eligible for the study is represented in the study.
5) Some potential participants may have not been included due to not having access to a computer, which was the predominant source of information used for recruitment.

6) Test-Retest was not completed with the instruments of this study. This questions the external consistency of the instruments.
7) The survey was only offered in English, possibly creating a sample bias.

Conclusion

Regardless of the limitations identified, this is the largest study of stress conducted with caregivers of individuals diagnosed with Prader-Willi syndrome. The results of this study suggest relationships between perceived social supports (negative) and two emotion-focused coping strategies (positive) are significant with stress for this population. Furthermore, the variables: hyperphagia, crisis cycle of maladaptive behaviors, resources/respites, and problem-focused coping strategies had no significant relationships with stress. Although this study could not support any strong or even moderate correlations between any of the independent variables with stress, it provided evidence that training in coping strategies could prove beneficial. Furthermore, results suggest that promotion of the use of social supports has been beneficial with the lowering of stress scores with this sample. The individuals showed lower stress levels with the increase of the use of social supports.

Resources and respite care still appears to be a major issue with these caregivers. Although their stress levels are shown to be lower than any other study, it appears they are doing it without the assistance of policy makers and professionals outside the field of PWS. They still are not receiving the assistance needed in proper daycare services and evening activities. It is not clear if the services are not being provided or they are being provided but they are not conducive to the supervision needs of individuals with PWS.

The findings of this study would be beneficial to caregivers as well as professionals working with individuals with PWS. Benefits could include 1) evidence to policy makers for the need of additional services for caregivers and individuals diagnosed with PWS and 2) the provision of interventions for caregivers in ways to prevent stress. Studies using longitudinal approaches could prove beneficial as well as research investigating mediating effects of the variables identified in this study.

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LIST OF APPENDICES

APPENDIX A

Appendix A: Pilot study email to participants

Dear Participants:

We would like to invite you to participate in a ***pilot*** research study on the relationship between caring for individuals diagnosed with Prader-Willi syndrome and caregiver stress: A Pilot Study. The results will provide vital information to caregivers, service providers, and policy makers on strategies and supports needed in the care of individuals with PWS. To participate, you must be at least 18 years of age and a family member who is a primary or secondary caregiver of an individual diagnosed with Prader-Willi syndrome. For this pilot study, you will be asked to provide suggestions or comments at the end of each section.

The survey will take approximately 30 minutes to complete. Because of the time commitment, at the end of the survey, all study participants, including pilot participants will have the opportunity to enter their name for a chance to receive one of eight $25 Amazon gift cards. In order to assure anonymity of your responses, at the end of this survey you will be directed to a separate survey form to enter the drawing. This ensures that there is no connection between your survey responses and your contact information.

All responses will be collected anonymously. Completion of this survey is completely voluntary. We value your participation in this study. By clicking the link below, you are agreeing to participate in this research study.

[insert survey link here]

This study has been reviewed by The University of Mississippi's Institutional Review Board (IRB). If you have any questions, concerns, or reports regarding your rights as a participant of research, please contact the IRB at (662) 915-7482 or irb@olemiss.edu.

Thank you,

Michael A. Vice
Doctoral Candidate, Health Behavior and Promotion
Department of Health, Exercise Science and Recreation Management
The University of Mississippi
Turner Center 234
662-832-0817
mavice@go.olemiss.edu

APPENDIX B

Appendix B: General study email to participants

Dear Participants:

We would like to invite you to participate in a research study on the relationship between caring for individuals diagnosed with Prader-Willi syndrome and caregiver stress. The results will provide vital information to caregivers, service providers, and policy makers on strategies and supports needed in the care of individuals with PWS.

The survey will take approximately 30 minutes to complete. Because of the time commitment, at the end of the survey, all study participants will have the opportunity to enter their name for a chance to receive one of eight $25 Amazon gift cards. In order to assure anonymity of your responses, at the end of this survey you will be directed to a separate survey form to enter the drawing. This ensures that there is no connection between your survey responses and your contact information.

All of your responses will be collected anonymously. Completion of this survey is completely voluntary. We value your participation in this study. By clicking the link below, you are agreeing to participate in this research study.
[insert survey link here]

This study has been reviewed by The University of Mississippi's Institutional Review Board (IRB). If you have any questions, concerns, or reports regarding your rights as a participant of research, please contact the IRB at (662) 915-7482 or irb@olemiss.edu.

Thank you,

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APPENDIX C

Appendix C: Questionnaire

This survey concerns individuals diagnosed with Prader-Willi syndrome and their caregivers. *Caregivers* are to be the individuals actually completing the survey. Your responses are anonymous. The survey should take approximately 30 minutes to complete and once the survey is complete, you will have the option of entering a drawing for one of eight $25 Amazon gift cards.

If more than one family member (caregiver) is participating, each person should complete a separate questionnaire and provide answers from his/her own perspective or opinion.

Note: Throughout the survey the term 'individual' references the individual with Prader-Willi syndrome that is being cared for.

For pilot study participants: At the end of each section there will be an opportunity to provide input for each question. Please provide any comments you feel may be beneficial for the implementation of this study. On the last section, please provide your input/opinion on the time it took to finish this survey.

Questionnaire:

Section One - Demographic Information

Instructions: The following questions are related to both you and the individual you care for. Please answer all questions to the best of your knowledge.

1. How old are you?
____ Under 18
____ 18-25
____ 26-34
____ 35-54
____ 55-64
____ 65 or over

2. Are you being paid to care for the individuals (i.e. direct care worker)?
____ Yes
____ No

3. What is the age of the individual you care for?

4. Does the individual you care for live in a supported living environment (i.e. supervised group home or supervised apartment)?
____ Yes
____ No

5. What is your relationship to the individual you care for?
____ Father ____ Mother
____ Stepfather ____ Stepmother
____ Grandfather ____ Grandmother
____ Sibling ____ Other

6. What type of primary residence does the individual you care for live in at present time?
____ Primarily lives in a group home/supportive living home
____ Primarily lives with you
____ Primarily lives with another family member or ex-family member
____ Equally shared residence with another family member or ex-family member
____ Other
____

7. What is your marital status?
____ Single
____ Married
____ Separated
____ Divorced
____ Widowed

8. What is your ethnicity?
____ African-American
____ Asian
____ Caucasian
____ Hispanic
____ Native-American
____ Other

9. What is the gender of the individual you care for?
____ Male ____ Female

10. What is the intellectual disability diagnosis (formerly known as mental retardation) of the individual you care for, if known?
_____ Unknown
_____ Average Range or above (85 or above)
_____ Borderline (between 70 – 84)
_____ Mild (IQ between 55 – 69)
_____ Moderate (IQ between 40 – 54)
_____ Severe (IQ between 25 – 39)
_____ Profound (IQ of less than 25)

11. What is the approximate height of the individual you care for?

12. What is the approximate weight in pounds of the individual you care for?

13. How often is access to food restricted while you are the caregiver (i.e. locked refrigerators, doors, pantries, and cabinets)?
____ Never
____ Rarely
____ Sometimes
____ Often
____ Always

14. What is the highest level of education you have completed?
____ Less than High School
____ High School/GED
____ Some College
____ 2-year College Degree
____ 4-year College Degree
____ Master's Degree
____ Doctoral Degree
____ Professional Degree (JD, MD)

15. How much do you feel the care you provide for individual prevents you from having gainful employment outside the home?
_____ Not particularly at all
_____ A little
_____ Somewhat
_____ Very much
_____ Extremely

16. What is your annual household income?
_____ Less than $25,000 per year
_____ Between $25,000 and $50,000 per year
_____ More than $50,000 and up to $75,000 per year
_____ More than $75,000 and up to $100,000 per year
_____ More than $100,000 per year

17. How many *other* children are living in the home?
_____ None
_____ One
_____ Two
_____ Three
_____ More than three

18. How many siblings are living outside the home?
_____ None
_____ One
_____ Two
_____ Three
_____ More than three

Section Two – Individual Care Needs/Resources &Respites (Adapted from Thomson, 2011)

Instructions: The following questions relate to the care needs of the individual with PWS. Please answer all questions to the best of your knowledge.

19. On average, how many hours per week do you spend caring for the individual at home and/or transporting?

_____ None
_____ At least one but less than ten hours per week
_____ At least ten but less than 20 hours per week
_____ At least 20 but less than 30 hours per week
_____ At least 30 but less than 40 hours per week
_____ 40 or more hours per week

20. On average, how many hours per week does the individual spend out of the home at school/employment/day center without you being present?

_____ None
_____ At least one but less than ten hours per week
_____ At least ten but less than 20 hours per week
_____ At least 20 but less than 30 hours per week
_____ At least 30 but less than 40 hours per week
_____ 40 or more hours per week

21. On average, how many hours per week does the individual you care for spend in leisure activities outside the residence without you being present (i.e. walking, team sports, shopping, bicycling, attending sporting activities and/or movies)?

_____ None
_____ At least one but less than ten hours per week
_____ At least ten but less than 20 hours per week
_____ At least 20 but less than 30 hours per week
_____ At least 30 but less than 40 hours per week
_____ 40 or more hours per week

22. Has the individual received respite care (inside or outside the home) within the last year? *Respite care is defined as temporary professional assistance enabling a break from caregiving of the individual.

_____ Yes _____ No

    23. If yes: On average, how many total days was respite care provided for the individual in the past year?

    _____ Up to one day within the past year
    _____ More than one day and up to 10 total days within the past year
    _____ More than 10 days and up to 20 total days within the past year
    _____ More than 20 days and up to 30 total days within the past year
    _____ More than 30 total days within the past year

24. On average, how many hours per day did you receive respite care in the past year?
_____ Less than one hour per day
_____ At least one hour and up to four hours per day
_____ More than four hours and up to eight hours per day
_____ More than eight hours and up to 16 hours per day
_____ More than 16 hours and up to 24 hours per day

25. How many out-of-home respite services have you used within the past year?
_____ One
_____ Two
_____ Three
_____ More than three

Section Three – Primary/Secondary Caregiver Health Status (Adapted from Lawton et al., 1982 and Thomson, 2011)

Instructions: The following questions are related to how you perceive your own health. Please answer all questions to the best of your knowledge.

26. Overall, my health status can be described as:
_____ extremely poor
_____ below average
_____ average
_____ below average
_____ excellent

27. In the past three years my health has:
_____ become much worse
_____ become a little worse
_____ stayed about the same
_____ become a little better
_____ become much better

28. Health problems prevent me from doing things I want to do:
_____ a great deal
_____ moderately
_____ slightly
_____ not at all
_____ I have no health problems

29. Compared to others my own age, my health is:
_____ extremely poor
_____ poor
_____ the same
_____ good
_____ excellent

Section Four – Caregiver Health Literacy (Chew et al., 2005)

Instructions: The following questions involve how well one understands medical-related terms. Please answer all questions to the best of your knowledge.

30. How often do you have someone help you read medical material?
_____ Always _____ Often _____ Sometimes _____ Occasionally

31. How confident are you in filling out medical forms?
_____ Always _____ Often _____ Sometimes _____ Occasionally

32. How often do you have problems learning more about medical conditions because of difficulty understanding written information?
_____ Always _____ Often _____ Sometimes _____ Occasionally

Section Five – Hyperphagia (Dykens, et al., 2007)

Instructions: The following questions are about food-related characteristics of the individual with Prader-Willi syndrome in your care. Please answer all questions to the best of your knowledge.

33. How upset does the individual generally become when denied a desired food?
____ Not particularly upset at all
____ A little upset
____ Somewhat upset
____ Very upset
____ Extremely upset

34. How often does the individual try to bargain or manipulate to get more food at meals?
____ A few times a year
____ A few times a month
____ A few times a week
____ Several times a week
____ Several times a day

35. Once the individual has food on his/her mind, how easy is it for you or others to re-direct the individual away from food to other things?
____ Extremely easy, takes minimal effort to do so
____ Very easy, takes just a little effort to do so
____ Somewhat hard, takes some effort to do so
____ Very hard, takes a lot of work to do so
____ Extremely hard, takes sustained and hard work to do so

36. How often does the individual forage/rummage through the trash for food (that you are aware of)?
____ Never
____ A few times a year
____ 1–2 times a month
____ 1–3 times a week
____ 4 to 7 times a week

37. How often does the individual get up at night to food seek (that you are aware of)?
____ Never
____ A few nights a year
____ 1 to 2 nights a month
____ 1 to 3 nights a week
____ 4 to 7 nights a week

38. How persistent is the individual in asking or looking for food after being told "no" or "no more"?
____ Lets go of food ideas quickly and easily
____ Lets go of food ideas pretty quickly and easily
____ Somewhat persistent with food ideas
____ Very persistent with food ideas
____ Extremely persistent with food ideas

39. Outside of normal meal times, how much time does the individual spend talking about food or engaged in food-related behaviors?
____ Less than 15 minutes a day
____ 15 to 30 minutes a day
____ More than 30 minutes and up to an hour a day
____ More than one hour and up to 3 hours a day
____ more than 3 hours a day

40. How often does the individual try to steal/sneak food or steal/sneak money/credit card to purchase food (that you are aware of?)
____ A few times a year
____ A few times a month
____ A few times a week
____ Several times a week
____ Several times a day

41. When others try to stop the individual from talking about food or engaging in food-related behaviors, it generally leads to:
____ No distress or upset
____ Mild distress or upset
____ Moderate distress or upset
____ Severe distress or upset
____ Extreme distress, behaviors can't usually be stopped

42. How clever or fast is the individual in obtaining food?
____ Not particularly clever or fast
____ A little clever or fast
____ Somewhat clever or fast
____ Very clever or fast
____ Extremely clever or fast

43. To what extent do food-related thoughts, talk, or behavior interfere with the individual’s normal daily routines, self-care, school, or work?
___ No interference
___ Mild interference; occasional food-related interference with normal daily routines, self-care, or work
___ Moderate interference; frequent food-related interference with normal daily routines, self-care, or work
___ Severe interference; almost daily food-related interference with normal daily routines, self-care, or work
___ Extreme interference, often unable to participate in normal daily routines, self-care, or work

44. How old was the individual when he/she first showed an increased interest in food?

45. How variable is the individual's preoccupation or interest in food?
___ Hardly ever varies
___ Usually stays about the same
___ Goes up and down occasionally
___ Goes up and down quite a lot
___ Goes up and down all the time

Section Six – Crisis Cycle of Maladaptive Behaviors

Instructions: The following questions are related to maladaptive behaviors which are often associated with Prader-Willi syndrome which create a crisis situation. A crisis in relation to these questions is defined as when the behavior is severe enough to seriously disrupt normal routine within the setting the individual is in. Examples of behaviors can be one or all of the following or comparable behaviors: extreme and persistent yelling or arguing, prolonged stubbornness, tantrumming, running away, threats of physical violence, aggression towards caregiver or others, destruction of property, and/or other signs that are threatening to the caregiver, others, or property, and/or unsafe for all involved.

46. How often does the individual's behavior reach a crisis?
____ Behaviors never reach a crisis
____ An average of once a year
____ An average of once every six months
____ An average of once a month
____ An average of once a week
____ Behaviors reach a crisis on a daily basis

47. Usually, when the individual is frustrated he/she…
____ does not become upset and stays in control of behavior
____ is mildly upset but stays in control of behavior.
____ is moderately upset but stays in control of behavior.
____ is extremely upset but stays in control of behavior.
____ is extremely upset and loses control of behavior (nonphysical: i.e. shouting, threatening, and/or relentless arguing, tantrumming, extreme stubbornness, threatening to run away).
____ becomes extremely upset and loses control of behavior (physical: i.e. running away, aggression and/or destruction).

48. How long does it usually take to become obvious the individual is upset?
____ Not applicable (individual has never reached a crisis stage)
____ Immediately
____ Not immediate but within a minute
____ More than a minute and up to five minutes
____ More than five minutes and up to ten minutes
____ More than ten minutes

49. After noticing the individual is upset, how long does it usually take for the situation to reach a crisis stage?
____ Not applicable (individual has never reached a crisis stage)
____ Immediately
____ Not immediate but within a minute
____ More than a minute and up to five minutes
____ More than five minutes and up to ten minutes
____ More than ten minutes

50. After reaching a crisis stage, how long does it usually take for the situation to peak?
____ Not applicable (individual has never reached a crisis stage)
____ Immediately
____ Not immediate but within a minute
____ More than a minute and up to ten minutes
____ More than ten minutes and up to 30 minutes
____ More 30 ten minutes

51. After the situation has peaked, how long does it usually take for the individual to show signs of calming down?
____ Not applicable (individual has never reached a crisis stage)
____ Less than five minutes
____ Between five minutes and 30 minutes
____ More than 30 minutes but less than two hours
____ Between two hours and five hours
____ More than five hours

52. After showing signs of calming down, how long does it usually take for the individual to completely calm down?
____ Not applicable (individual has never reached a crisis stage)
____ Less than five minutes
____ Between five minutes and 30 minutes
____ More than 30 minutes but less than two hours
____ Between two hours and five hours
____ More than five hours

53. On the average, how pleasant is the environment in your home prior to any crisis being noticed?
____ Not applicable, (individual has never reached a crisis stage)
____ Extremely pleasant
____ Somewhat pleasant
____ A little pleasant
____ Not very pleasant
____ Not pleasant at all

54. On the average, how unpleasant is the environment in your home during a crisis?
____ Not applicable, (individual has never reached a crisis stage)
____ Not unpleasant at all
____ A little unpleasant
____ Very unpleasant
____ Extremely unpleasant
____ Almost intolerable

55. On the average, how long does it take for the environment to get back to normal after a crisis situation has ended?

____ Not applicable, (individual has never reached a crisis stage)
____ Less than an hour
____ Between one and five hours
____ More than five hours and up to ten hours
____ More than ten hours and up to 24 hours
____ Over 24 hours

Section Seven: Brief COPE (Carver, 1997)

Instructions: These items deal with ways you've been coping with the stress related to the caregiving of an individual diagnosed with PWS. People use a variety of different ways to deal with problems. These questions ask what you've been doing to cope with behavioral issues associated with PWS. Each item says something about a particular way people cope with stressful situations. The Following questions only ask if you are doing what is asked, not if it works. Please try to answer each question individually without considering the preceding or later questions.

56. I've been turning to work or other activities to take my mind off things.

____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

57. I've been concentrating my efforts on doing something about the situation I'm in.

____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

58. I've been saying to myself "this isn't real."

____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

59. I've been using alcohol or other drugs to make myself feel better.

____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

60. I've been getting emotional support from others.

____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

61. I've been giving up trying to deal with it. .
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

62. I've been taking action to try to make the situation better.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

63. I've been refusing to believe that it has happened.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

64. I've been saying things to let my unpleasant feelings escape.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

65. I’ve been getting help and advice from other people.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

66. I've been using alcohol or other drugs to help me get through it.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

67. I've been trying to see it in a different light, to make it seem more positive.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

68. I’ve been criticizing myself.
____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

69. I've been trying to come up with a strategy about what to do.
____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

70. I've been getting comfort and understanding from someone.
____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

71. I've been giving up the attempt to cope.
____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

72. I've been looking for something good in what is happening.
____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

73. I've been making jokes about it.
____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

74. I've been doing something to think about it less, such as going to movies, watching TV, reading, daydreaming, sleeping, or shopping.
____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

75. I've been accepting the reality of the fact that it has happened.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

76. I've been expressing my negative feelings.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

77. I've been trying to find comfort in my religion or spiritual beliefs.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

78. I’ve been trying to get advice or help from other people about what to do.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

79. I've been learning to live with it.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

80. I've been thinking hard about what steps to take.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

81. I’ve been blaming myself for things that happened.
_____ I haven't been doing this at all
_____ I've been doing this a little bit
_____ I've been doing this a medium amount
_____ I've been doing this a lot

82. I've been praying or meditating.
____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

83. I've been making fun of the situation.
____ I haven't been doing this at all
____ I've been doing this a little bit
____ I've been doing this a medium amount
____ I've been doing this a lot

Section Eight: Social Provisions Scale (Russell & Carolyn Cutrona, 1984)

Instructions: In answering the following questions, think about your current relationships with friends, family members, co-workers, community members, and so on. Please indicate to what extent each statement describes your current relationships with other people.

84. There are people I can depend on to help me if I really need it.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

85. I feel that I do not have close personal relationships with other people.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

86. There is no one I can turn to for guidance in times of stress.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

87. There are people who depend on me for help.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

88. There are people who enjoy the same social activities I do.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

89. Other people do not view me as competent.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

90. I feel personally responsible for the well-being of another person.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

91. I feel part of a group of people who share my attitudes and beliefs.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

92. I do not think other people respect my skills and abilities.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

93. If something went wrong, no one would come to my assistance.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

94. I have close relationships that provide me with a sense of emotional security and well-being.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

95. There is someone I could talk to about important decisions in my life.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

96. I have relationships where my competence and skill are recognized.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

97. There is no one who shares my interests and concerns.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

98. There is no one who really relies on me for their well-being.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

99. There is a trustworthy person I could turn to for advice if I were having problems.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

100. I feel a strong emotional bond with at least one other person.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

101. There is no one I can depend on for aid if I really need it.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

102. There is no one I feel comfortable talking about problems with.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

103. There are people who admire my talents and abilities.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

104. I lack a feeling of intimacy with another person.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

105. There is no one who likes to do the things I do.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

106. There are people who I can count on in an emergency.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

107. No one needs me to care for them.
____ Strongly disagree
____ Disagree
____ Agree
____ Strongly Agree

Section Nine: Mental Health Depression, Anxiety, Stress Scale (DASS 21)

In answering the following questions, please read each statement and choose the answer which indicates how much the statement applied to you over the past few weeks. There are no right or wrong answers. Do not spend too much time on any statement.

108. I found it hard to wind down.
____ Never
____ Sometimes
____ Often
____ Almost Always

109. I was aware of dryness of my mouth.
____ Never
____ Sometimes
____ Often
____ Almost Always

110. I could not seem to experience any positive feeling at all.
____ Never
____ Sometimes
____ Often
____ Almost Always

111. I experienced breathing difficulty (e.g. excessively rapid breathing, breathlessness in the absence of physical exertion).

Never
____ Sometimes
____ Often
____ Almost Always

112. I found it difficult to work up the initiative to do things.
____ Never
____ Sometimes
____ Often
____ Almost Always

113. I tended to over-react to situations.
____ Never
____ Sometimes
____ Often
____ Almost Always

114. I experienced trembling (e.g. in the hands).
____ Never
____ Sometimes
____ Often
____ Almost Always

115. I felt that I was using a lot of nervous energy.
____ Never
____ Sometimes
____ Often
____ Almost Always

116. I worried about situations in which I might panic and make a fool of myself.
____ Never
____ Sometimes
____ Often
____ Almost Always

117. I felt that I had nothing to look forward to.
____ Never
____ Sometimes
____ Often
____ Almost Always

118. I found myself getting agitated.
____ Never
____ Sometimes
____ Often
____ Almost Always

119. I found it difficult to relax.
____ Never
____ Sometimes
____ Often
____ Almost Always

120. I felt down-hearted and blue.
____ Never
____ Sometimes
____ Often
____ Almost Always

121. I was intolerant of anything that kept me from getting on with what I was doing.
____ Never
____ Sometimes
____ Often
____ Almost Always

122. I felt I was close to panic.
____ Never
____ Sometimes
____ Often
____ Almost Always

123. I was unable to become enthusiastic.
____ Never
____ Sometimes
____ Often
____ Almost Always

124. I felt I was not worth much as a person.
____ Never
____ Sometimes
____ Often
____ Almost Always

125. I felt that I was rather touchy.
____ Never
____ Sometimes
____ Often
____ Almost Always

126. I was aware of the action of my heart in the absence of physical exertion (e.g. sense of heart rate increase, heart missing a beat).
____ Never
____ Sometimes
____ Often
____ Almost Always

127. I felt scared without any good reason.
____ Never
____ Sometimes
____ Often
____ Almost Always

128. I felt that life was meaningless.
____ Never
____ Sometimes
____ Often
____ Almost Always

CURRICULUM VITAE

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ACADEMIC RECORD

Doctor of Philosophy, 2017 (Anticipated Graduation)
Major Area: Health and Kinesiology
Program: Health Behavior and Promotion
Department of Health, Exercise Science & Recreation Management
School of Applied Sciences
University of Mississippi
Dissertation: The Relationship Between Caring for Individuals Diagnosed with Prader-Willi Syndrome and Caregiver Stress

Education Leadership Certification, 1997
Major Area: Education Leadership
Department of Leadership and Counselor Education
School of Education
University of Mississippi

Master of Science in Education, 1993
Major Area: Emotional Disabilities
Department of Special Education
School of Education and Psychology
University of Southern Mississippi

Bachelor of Science in Recreation, 1983
Major Area: Recreation Administration
Department of Recreation, Park, and Tourism Studies
School of Public Health
Indiana University

PUBLICATIONS

Peer-reviewed

Vice, M.A., Nahar, V.K., Ford, M.A., Bass, M., Johnson, A.K., Davis, A.B., & Biviji-Sharma, R. (2015). Risk factors for low bone mineral density in institutionalized individuals with developmental disabilities. *Health Promotion Perspectives*, *5*(2), 147-152.

Nahar, V.K., **Vice, M.A.**, & Ford, M.A. (2013). Conceptualization and measuring risk perceptions of skin cancer: A review. *California Journal of Health Promotion*, 11(3), 36-47.

Nahar, V.K., Ford, M.A., Hallam, J.S., Bass, M.A., & **Vice, M.A.** (2013). Socio-demographic and psychological correlates of sun protection behaviors among outdoor workers: A review. *Journal of Skin Cancer*, 1-10.

Nahar, V.K., Ford, M.A., Hallam, J.S., Hutcheson, A., & **Vice, M.A.** (2013). Skin cancer knowledge, beliefs, self-efficacy, and preventive behaviors among North Mississippi landscapers. *Dermatology Research and Practice*, 1-7.

PRESENTATIONS

National Level

Ford, M. A., Nahar, V. K., Bass, M. A., **Vice, M. A.**, Davis, R. E., & Hutcheson, A. “Influences on bone mineral density among Asian Indians Residing in the US.” American College of Sports Medicine, Annual Meeting, Orlando, FL, May - June, 2014.

Vice, M. A., Nahar, V. K., Ford, M. A., & Bass, M. “Risk Factors for Low Bone Mineral Institutionalized Individuals with Developmental Disabilities.” American College of Sports Medicine, Annual Meeting, San Francisco, CA, May - June, 2012.

PROFESSIONAL CONFERENCES/MEETINGS ATTENDED

American College of Sports Medicine, Annual Meeting, San Francisco, CA, 2012

HIGHER EDUCATION TEACHING EXPERIENCE

Co-Instructor:

HP191 – Personal and Community Health

Guest Instructor/Speaker: Undergraduate School Level, University of Mississippi

EDSP 308 - Introduction to Special Education

DSP 329 - Nature and Needs of Children with Severe Disabilities

PSY 201 - General Psychology

PSY 301 - Developmental Psychology

EDSP 335 - Assessment of Exceptional Students

PROFESSIONAL EXPERIENCE

Dec 2016 - Present	***Cultural/Linguistic Competency Coordinator (Contractual)*** ***(Communicare Region II Mental Health)*** Addressing disparities of a four-county region in mental health services and ensuring all children with severe emotional disabilities and family members have access to the provided services. Assisting with establishing policies and procedures, and designing new programs which meet the needs of consumers using a logic model. The services are provided to enable high-risk children to live in their natural settings rather than institutions. Evaluation of existing programs to assess cultural/linguistic competency and make recommendations to administration.
July 2016 - Dec 2016	***Retired/Career Transition*** Pursuing a second career in Higher Education.
July 2005 - June 2016	***Principal/Special Education Director*** ***(North Mississippi Regional Center: Oxford, MS)*** Overall supervision, direction, and training of the Special Education Department covering 23 northern Mississippi counties and 80 employees. Staffing includes: assistant principal, workshop director, teachers, assistant teachers, speech pathologists; audiologists and an early intervention evaluation team. A GED program was offered periodically to interested individuals.

Professional Experience (continued)

April 2001 - June 2005	***Department Director: Residential Services*** ***(North Mississippi Regional Center: Oxford, MS)*** Overall supervision, direction, and training of 300 staff within the Resident Services Department. Staffing included: direct care staff for 12 residential cottages, beauty shop/parlor, sewing room, janitorial crew, supplies, and student transportation.
Jan 2001 - March 2001	**Department Director: Advocacy Services** ***(North Mississippi Regional Center: Oxford, MS)*** Responsibilities included: Ensuring adherence to (and reporting to) all local, state, and federal regulations regarding client protection from abuse, neglect, and/or exploitation; formulating, directing, and managing the operations of a division through subordinate personnel.
Nov 1994 - Dec 2000	**Assistant Personnel Director: Personnel Services** ***(North Mississippi Regional Center: Oxford, MS)*** Assisted Personnel Director with all personnel matters for staffing of over 1000 employees, covering 23 northern Mississippi counties: Abuse Investigator; Drug and Alcohol Testing Coordinator: Quarterly Blood Drive Coordinator.
Nov 1993 - Oct 1994	***Masters Level Teacher*** ***(North Mississippi Regional Center: Oxford, MS)*** Teacher of 25 adult students with intellectual/developmental disabilities ranging from mild to severe, many of which had secondary diagnoses of various behavioral /emotional disabilities and medical diagnoses.
Nov 1991 - Oct 1993	***Staff Development Instructor*** ***(Ellisville State School: Ellisville, MS)*** Instructed all new employees and veteran employees of facility in: Policies and Procedures; local, state, and federal regulations. Courses taught included, but not limited to, CPR, First Aid, Techniques in Managing Aggressive Behaviors (TMAB), Teachers' Inservice, Campus-Wide Monthly Inservices, Direct Care Worker Upgrade class, Reading courses for staff and students of facility. Coordinated and organized one-to-three day workshops.
June 1990 - Oct 1991	***Recreation Supervisor*** ***(Ellisville State School: Ellisville, MS)*** Provided recreational and physical activities for over 50 students diagnosed with mild to severe intellectual/developmental disabilities, many of which had secondary diagnoses of various behavioral /emotional disabilities and medical diagnoses.

CERTIFICATIONS/LICENSE

Mississippi Department of Education Certified School Administrator

Mississippi Department of Education Certified AA Teacher (ED)

Tennessee Department of Education Certified School Administrator

Tennessee Department of Education Certified Teacher

Mississippi Certified Public Manager

Mississippi Certified Public Supervisor

Mississippi Certified Mental Health/Intellectual and Developmental Disability Administrator

Mississippi Certified Intellectual/Developmental Disability Therapist

Mississippi Certified Mental Health Therapist

Mississippi Stennis Institute Graduate

Millsap’s College Principals’ Summer Institute Graduate

Mississippi Autism Task Force State trainer

Mandt Behavioral Intervention trainer

American Heart Association CPR instructor

Online Teaching and Learning trained

Collaborative Institutional Training Initiative (CITI)