Caregiver Reactions in Pulmonary Hypertension

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Chapter 1: Introduction

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Abstract

This dissertation contains a modified Walker and Avant (2005) concept analysis on caregiver reactions, a review of empirical referents, and a qualitative phenomenological investigation into informal caregiver reactions in pulmonary hypertension (PH). In the second chapter, the assumption of burden in caregiving is a presupposition, so the importance of bracketing and inclusion of positive reactions in caregiving is highlighted in the concept analysis. This chapter further identified and determined the defining attributes of informal caregiver reactions, as well as antecedents and consequences. A model case is also presented. Chapter three reviews the Bakas Caregiving Outcomes Scale (BCOS), the Caregiver Reaction Assessment (CRA), the Modified Caregiver Strain Index (MCSI), the Zarit Burden Interview (ZBI), the Perceived Caregiver Burden Scale (PCBS), the Multidimensional Caregiver Burden Inventory (MCBI), and the Screen for Caregiver Burden (SCB) (Bakas, Champion, Perkins, Farran, & Williams, 2006; Given et al., 1992; Novak & Guest, 1989; Stommel, Given, & Given, 1990; Thornton & Travis, 2003; Vitaliano, Russo, Young, Becker, & Maiuro, 1991; Zarit, Reever, & Bach-Peterson, 1980). These instruments are investigated for reliability and validity, conceptual frameworks, and use of instruments in the literature. The fourth chapter consists of a qualitative study of informal caregivers of those with PH. A semi-structured interview was utilized, and six overarching themes were found. They included Initial Diagnosis Overwhelming, Becoming Clinically Literate, Embracing A New Normal, Caregiving Creates Stress, Getting Support and Self-Care, and Improving Relationship Quality.

Keywords: caregiver burden, informal caregivers, caregiver reactions, concept analysis, psychometrics
The informal caregiver is a paramount consideration in the care of not only a person with pulmonary hypertension (PH), but also to any individual requiring care. In the United States, there are an estimated 43.5 million caregivers over 18 years of age who provide unpaid care to an adult family member. A typical caregiver provides an average of 24.4 hours of care per week. The rate of caregiving is increasing, with an estimated prevalence of 16.6% of American adults caring for another adult (American Association of Retired Persons [AARP], 2015). Specifically with PH, the caregiving role is vital to the health of the care receiver and the overall severity of disease makes the phenomenon an important one to study (Flattery, Pinson, Savage, & Salyer, 2005; Hoeper, 2009; Matura, McDonough, & Carroll, 2016; Steinbis, 2008). Caregiving can potentially introduce strain into multiple areas of life, including the caregiver’s own health, finances, time availability, family support, and employment (Given, Given, Stommel, Collins, King & Franklin, 1992). Conversely, there are positive aspects of caregiving, such as positive self-esteem, improvement of relationship, role satisfaction, and pride. Positive aspects of caregiving are an important consideration because without inclusion of the positive, the entire phenomenon would not be captured. Assuming solely the presence of burden leads to researcher bias, and an invalid capturing of the caregiving experience.

PH disease process is a chronic, progressive illness that arises from a mean pulmonary artery pressure of greater than or equal to 25mm Hg at rest, measured by a right heart catheterization (Hoeper et al., 2013). The increased pulmonary artery pressure causes such symptoms as dyspnea, pain, anxiety, cyanosis, dizziness, syncope, and edema (Matura & Carroll, 2010). There is a paucity of literature about the caregiver within the context of PH, and the symptom-related quality of life research of the care receiver is still in its infancy. It is important to add to the knowledge base about informal caregiving in PH, as investigating
informal caregiving is a priority of the National Institute of Nursing Research (NINR) (NINR, 2011). The severity of the disease, cost of treatment, as well as seriousness of medications calls for investigation into the caregivers’ experiences. Social support has been demonstrated to increase health-related quality of life (Leung, Pachana, & McLaughlin, 2014), implying that informal caregivers play an important role in their caregiver’s well-being.

In the second chapter, the conceptual basis and history of the literature regarding “burden” is explored utilizing a modified Walker and Avant (2005) concept analysis approach. “Burden” typically has a negative connotation in both literature and everyday diction, but this negative concept is what has been traditionally addressed in the literature. The evolution of the multidimensional nature of caregiver reaction literature is discussed, as well as suggestions for terminology and bracketing, or decreasing bias when going forward with research involving caregiver reaction. Implications and significance for informal caregivers also is discussed, as this is an important topic following trends in deinstitutionalization, shortening hospital lengths of stay, chronic care in the home, and adult-child caregivers.

Psychometric instruments are pivotal in the objective measurement of a given phenomenon. Chapter three addresses measurements of informal caregiver reactions in an investigation into empirical referents. “Caregiver reactions” is an elusive concept to capture and measure, but multiple instruments are available for use. This chapter provides a review of such instruments enabling a smooth transition from qualitative to quantitative measurement in the PH caregiver. The instrument review also fulfills the empirical referents portion of the preceding Walker and Avant (2005) concept analysis. These instruments are compared for the specific phenomenon measured; and validity and reliability are also discussed. The instruments reviewed include Bakas Caregiving Outcomes Scale (BCOS), the Caregiver Reaction Assessment (CRA),
the Modified Caregiver Strain Index (MCSI), the Zarit Burden Interview (ZBI), the Perceived Caregiver Burden Scale (PCBS), the Multidimensional Caregiver Burden Inventory (MCBI), and the Screen for Caregiver Burden (SCB) (Bakas, Champion, Perkins, Farran, & Williams, 2006; Given et al., 1992; Novak & Guest, 1989; Stommel, Given, & Given, 1990; Thornton & Travis, 2003; Vitaliano, Russo, Young, Becker, & Maiuro, 1991; Zarit, Reever, & Bach-Peterson, 1980).

The strengths and weaknesses of each instrument are assessed, as there is not necessarily one “best” psychometric instrument. Inclusion of positive reactions to caregiving within each instrument is also explored.

Informal caregivers are invaluable in their role in the PH dyad. The purpose of the project presented in Chapter 4 was to describe the experience of the informal PH caregiver. Specific aims included producing a rich, non-biased description of this specific caregiving experience. For the most comprehensive description of the phenomenon, a semi-structured, open-ended interview guide was utilized, with analysis using Moustakas’ modification of the Stevick-Colaizzi-Keen method of phenomenological investigation, with two-cycle coding (In Vivo and Process coding; followed by thematic organization) (Saldaña, 2009). Participants were recruited from the Boston PH support group and the Pulmonary Hypertension Association’s online message board.

The concept analysis on caregiver burden, the investigation of empirical referents, and this novel research on informal caregiver reactions in PH effectively call for further research into the PH caregivers’ experiences. The informal caregiver is an extremely important part of the care receiver’s overall experience and health-related quality of life, making this group imperative to study.
References


Chapter 2: The Caregiver Experience and Presence of Burden: A Concept Analysis

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Abstract
This modified Walker and Avant (2005) concept analysis investigates informal caregiver burden and the history of the literature involving burden. Avoiding bias and bracketing is highlighted in word choice involving burden, as it supposes negative reactions to caregiving. The inclusion of positive reactions to caregiving was not incorporated in the literature until the early 1990s, but this emphasizes the importance of using more neutral diction in caregiver reaction research. The following article aims to identify and determine the defining attributes of informal caregiver reactions. Additionally, antecedents and consequences are explored, which are events that must occur prior to the manifestation of a concept, and events that occur as a result of a concept, respectively. Lastly, a model case of the concept is presented.

*Keywords:* caregiver burden, informal caregivers, caregiver reactions
The Caregiver Experience and Presence of Burden: A Concept Analysis

Caregiver burden is a concept that is difficult to define and analyze, as the word implies a negative connotation, making bracketing presuppositions very challenging. “Caregiver” in this usage could be defined as either a lay or medically trained professional. For the purposes of this paper, the primary focus will be the untrained, informal, or lay caregiver. “Burden” and caregiving experiences are the primary foci of this analysis. “Burden,” is defined by the Merriam-Webster dictionary as “something that is carried; a duty, responsibility; something oppressive or worrisome” (Merriam-Webster’s Dictionary, 2016). Formal accepted nomenclature is important to establish as part of the concept analysis. The concept of a caregiver’s “experience” is an important consideration; positive aspects of caregiving are present, albeit not well studied. Use of the word “experience” presents a more neutral term to capture the extent of a caregiver’s involvement. The purpose of this paper is to perform a Walker and Avant (2005) concept analysis on the caregiver experience and burden, which will be slightly modified to identify the concept, determine the defining attributes, or noting characteristics that repeat in the literature. Antecedents and consequences will be discussed; that is, the conditions that must be in place for burden to exist, and results of the concept, respectively. Implications and significance of burden will be discussed, in addition to suggestions for terminology in qualitative research.

Evolution of Caregiver Burden Research

A Two-Dimensional Concept

There is still much disagreement in the literature regarding the conceptual and operational definitions of burden. Many researchers have argued that burden should only refer to subjective
perceived stress or strain in the context of caregiving (Stommel, Given, & Given, 1990), but objective burden still remains important, as well as positive reactions within the caregiver’s experience. It is important to remember that burden is a multidimensional concept; objective measures of burden (e.g. time, money, energy spent caregiving) are irrevocably linked to the overall perception of caregiving experience. Objective burden, as part of the overall experience, is important to include in a conceptual definition of the caregiver’s experience.

In caregiver experience research, the caregiver is defined as an unpaid individual who assists in the care of a person with a self-care deficit (Hunt, 2003). Burden, or a response to perceived stress or negative assessment (e.g. oppression) to taking care of a person or people who are ill, was introduced in the mid-1960s as a largely two-dimensional concept. Burden had an objective (e.g. financial) and subjective (e.g. emotional) component, all seemingly negative as one could surmise from the connotation of the word (Bastawrous, 2013; Kim, Chang, Rose, & Kim, 2012). Burden is a multifaceted phenomenon. It was initially introduced by Hoenig and Hamilton as a dichotomous concept consisting of objective and subjective burden as early as the late 1960s (Hoenig & Hamilton, 1966). These authors suggested that the physical and financial or more objective dimensions of burden should be separated from the emotional response, reinforcing this concept of burden as dichotomous. Although this study was the most in-depth of its time, burden as a multidimensional concept was not substantiated.

**Gerontology Related Research: Caregiving as Multidimensional**

Caregiver burden was largely studied in gerontology and psychology in the late 1970s to early 1980s, with most dependent variables in the literature being activities of daily living (ADLs) and social support (Chappell & Funk, 2011). In this period of early caregiver burden research, sole focus was on observable as opposed to perceived strain (Hunt, 2003). Later,
caregiver burden was defined by George and Gwyther (1986) as incorporating the physical, psychological, emotional, social, and financial stresses of caregiving as subcategories to the subjective and objective components, qualifying burden as multidimensional (Bastawrous, 2013; George & Gwyther, 1986). Zarit et al. (1980; Zarit et al., 1986) studied and defined caregiver burden, contributing the most widely accepted conceptual definition of burden, as well as extrapolating the multifaceted nature of burden. Zarit later developed the seminal Zarit Burden Interview (ZBI) to help measure the concept of burden (Zarit, Reever, & Bach-Peterson, 1980; Zarit, Todd, & Zarit, 1986). This was the first study to include a spiritual domain to caregiving. The authors also introduced the concept of caregiving being a unique experience, with the ability to be emotionally rewarding in addition to carrying potential for perceived stress. Although this positive response to caregiving was only briefly reviewed, it directed researchers in the early 1990s to further investigate the concept of positive aspects of caregiving. Zarit et al. defined caregiving burden as “the extent to which caregivers perceive that caregiving has had an adverse effect on their emotional, social, financial, physical, and spiritual functioning” (Sielger, Capezuti, Mezey, 2007, pp. 111; Zarit et al., 1986), successfully capturing a conceptual definition while still incorporating all aspects of both the subjective and objective aspects of caregiver burden. Despite this success, they do not incorporate the positive aspects of caregiving into their conceptual definition from 1980, but do include them in the ZBI instrument validation study published in 1986 (Zarit et al., 1980; Zarit et al., 1986). Zarit et al. (1980, 1986) were the first to shift focus beyond the objective domains, a novel idea for the time (Zarit et al., 1980). Additionally, Montgomery et al. (1985) studied this phenomenon as a dynamic view of the caregiving dyad which should not be limited to the dependent elderly, opposing Zarit’s extensive study of an older population with Alzheimer’s disease. They also emphasized that there were
subjective and objective measures and sentiments in burden (Montgomery, Gonyea, & Hooyman, 1985).

A Shift to the Positive

By 1990, research on informal caregiving changed dramatically, as attention shifted to the large and aging baby boomer generation, and more funding was provided to institutions supporting gerontological research (Chappell & Funk, 2011). Caregiver burden continued to be almost exclusively investigated as a negative concept until the early 1990s, when Given et al. (1992) introduced the more neutral term “reaction,” allowing for measurement of a caregiver esteem domain—described by the authors as a sense of pride derived from caregiving (Given et al., 1992). This is an important paradigm shift because it incorporates an entire domain of caregiving that was otherwise formerly disregarded. Given et al. also included family support, finances, schedule, and caregiver health as domains, thus expanding the dichotomy of subjective and objective burden into multiple domains (Given et al., 1992). This newly accepted view of burden as multidimensional helps to eliminate bias in ongoing research on caregivers.

Interestingly, Stommel and Given (1990) choose to only investigate the subjective perceived measures of burden; they asserted that the objective domain could potentially cloud the subjective perception of burden (Stommel et al., 1990, pp. 83). Contrary to this, Given’s 1992 instrument clearly incorporates objective measures integral to the caregiver’s overall experience. The summary of the evolution of burden literature demonstrates important points: “caregiver experience” would be the most correct diction to utilize in research to understand the full phenomenon, as “burden” supposes only negative reactions. Caregiver burden and experience are multidimensional concepts that should not be separated and reduced to solely subjective and objective domains. Investigation into the full caregiver’s experience requires bracketing to avoid
bias in qualitative research, as one should not assume the presence of burden. Terms like caregiver “experience”, “response”, or “reactions” would help to reduce presuppositions by the researcher. Additionally, journaling can be utilized to identify and avoid potential bias by assuming presence of burden.

**Defining Attributes and Antecedents: The Informal Caregiver’s Experience**

Defining attributes are properties that delineate one concept from another to clarify its meaning (Walker & Avant, 2005). Defining attributes of the caregiver’s experience have been identified as (1) the ability to provide care for another individual, (2) recognition that the provision of care elicits a response, and (3) the expression of the caregiver’s response—negative or positive—as well as any ensuing reactions. For informal caregiver burden to exist, there first must be antecedents, or conditions present prior to the occurrence of burden or the caregiver’s experience (Walker & Avant, 2005). With the informal caregiver experience, there is an individual who requires the care of a person possessing no background in medicine or nursing, and this individual is unpaid. Familial relation to care receiver is irrelevant if the caregiver is providing moderately frequent care to the receiver. The relationship must not be coerced, and the caregiver must be involved in the care receiver’s direct physical and emotional support, assistance in activities of daily living (ADLs), finances, household errands, and shopping, as well as any action a caregiver would deem as falling under providing care for their care receiver.

**Consequences: Global Scope of Caregiver Burden**

Assistance for caregivers is a topic discussed in the National Institute of Nursing Research (NINR) strategic plan, as well as self-management of chronic disease (NINR, 2016).
With an aging population and need for more cost-effective out-of-facility care, incidence of informal caregiving burden will increase. There is an unprecedented growth in number of persons ages 65 and over, currently at 89 million people. This population is projected to double by 2050 (Centers for Disease Control and Prevention, 2013). Additionally, the number of people with chronic disease is also rapidly increasing, with 133 million Americans, roughly 45% of the population, having at least one chronic disease. As of 2012, about half of all adults in the United States has one or more chronic health conditions (Ward, Schiller, & Goodman, 2012). Those with chronic diseases are frequent users of health care. They account for 81% of hospital admissions, 91% of all prescriptions filled, and 76% of all physician visits (Anderson, 2004).

With the incidence of chronic disease increasing, cost of care will invariably increase unless there are reasonable interventions enabling self-efficacy for the care receiver and support for the informal caregiver. In fact, chronic disease accounted for 86% of all health care spending in 2010, with heart disease and stroke costing an estimated $315.4 billion, and cancer costing $157 billion (Gerteis et al., 2014; Go et al., 2014; Mariotto, Yabroff, Shao, Feuer, & Brown, 2011). Promoting a program of self-efficacy for the care receiver has shown to decrease hospitalization costs an average of $2,149 (2004 Canadian dollars) per patient, with an estimated savings of about $1,300 per prevented hospital admission (Bourbeau et al., 2006). Opportunity costs of informal caregiving of elderly adults have amounted to $522 billion annually, with an estimate of 22-26 billion hours of caregiving given annually (Chari, Engberg, Ray, & Mehrotra, 2015).

**Consequences Continued: Implications for the Caregivers in the Context of Various Disease States**
Heart Failure

Heart failure (HF) literature includes a multitude of evidence pertaining to self-efficacy, or an individual’s perception of his or her capacity to complete tasks related to maintenance of health or environment, a concept studied by Bandura in the late 1970s (Bandura, 1977). When there are threats to self-efficacy, the informal caregiver plays an important role in provision of care. This concept has been investigated in the context of HF. A study of 135 partners of HF patients by Agren et al. (2010) utilized the Caregiver Burden Scale. They found that 30% of the caregivers perceived burden as medium, and 2% reported burden as high. The sample of HF care receivers were New York Heart Association (NYHA) functional class II-III HF patients, indicating lower requirements of at-home care in comparison to stage IV individuals who are symptomatic at rest. Examining these functional class IV individuals would likely reveal greater burden. The most difficult area for caregivers was ability to have some control over cardiac care of their partner, as well as maintaining their own freedom and social interaction. (Ågren, Evangelista, & Strömberg, 2010). Further study on HF caregivers demonstrated that if caregivers had a higher perceived burden, there was a perception of more time spent on caregiving tasks, as well as a higher rate of anxiety and poorer health-related quality of life of the caregiver. These trends tend to generally decrease over time (Pressler et al., 2013). Despite the decrease, caregiver burden is interconnected to the care receiver’s health and well-being, care receivers’ levels of depression increased with higher level of perceived caregiver burden among the caregivers. Caregiver’s depression was also positively correlated with their care receiver’s depression, as well as with lower perceived social support, poor relationship quality, and decreased confidence in HF management (Trivedi, Piette, Fihn, & Edelman, 2012). Also pertaining to burden in HF, a smaller exploratory study found relationship quality positively
correlated with caregiver benefit and negatively correlated with burden and depression (Lum, Lo, Hooker, & Bekelman, 2014), indicating the importance of relationship quality in the caregiving dyad. A qualitative inquiry by Gysles and Higginson (2009) encompassed multiple disease states and caregiver’s experiences, including caregivers of those with heart failure, chronic obstructive pulmonary disease (COPD), motor neuron disease, and/or cancer. The authors found that caregivers were not very well equipped to deal with severe shortness of breath in the home setting.

Social support, support from healthcare professionals, and further education are important considerations for the HF caregiver. Needs expressed by HF dyads included: a need to share burden, increased guidance through the illness trajectory, assistance in management of daily living, increased social support (both via dyadic support as well as family/friends/others), and increased access to telephone support (Graven & Grant, 2014; Liljeroos, Ågren, Jaarsma, & Strömberg, 2014; Lyons et al., 2015). They also report limited knowledge about end of life, and changing needs of the care receiver- including declining functions and safety, specifically early in the HF disease trajectory (Bekelman et al., 2011; Laoruangroch, Nauser, & Warmbrodt, 2013). Caregivers specifically need further support from clinicians, to be not only involved in care, but to have clearly defined and involved roles. Without this support, caregivers displayed anger and distrust in the health care system (Burke, Jones, Ho, & Bekelman, 2014). Additionally, family support and the caregiver’s own health is vital to the care receiver (Boyoung, Fleischmann, Howie-Esquivel, Stotts, & Dracup, 2011).

**Alzheimer’s Disease**

Alzheimer’s disease (AD), in which care can be particularly burdensome, offers a different perspective from HF. Much of the AD literature targets the adult child as primary
caregiver. The adult child perceives greater amounts of burden in comparison to a spousal relationship in the context of AD, which does not decrease over time as seen in spousal relationships (Chappell, Dujela, & Smith, 2014). In one study with a particularly large sample size of 1,235 AD patients from Spain, caregiver burden decreased in association with increasing AD patients’ education levels. Higher education levels among caregivers have also been associated with decreased burden, including lower incidence of depression and fewer behavioral symptoms in the patients. AD patients’ behavioral symptoms contributed significantly to burden levels of caregivers (Agüera-Ortiz, Frank-García, Gil, & Moreno, 2010), as well as difficulty in medication management (Smith, Grijseels, Ryan, & Tobiansky, 2015). Further confirming presence of burden, a study by Bergvall et al., (2011) found that severity of behavioral disturbances was the strongest predictor of caregiver burden using the Zarit Burden Interview. Ability to perform activities of daily living (ADLs) was the strongest predictor of informal care hours (Bergvall et al., 2011). Further studies demonstrate significantly increased burden of care in AD (Brodaty et al., 2014; D’Onofrio et al., 2015; Miller, Rosenheck, & Schneider, 2012; Raggi, Tasca, Panerai, Neri, & Ferri, 2015; Yeager, Hyer, Hobbs, & Coyne, 2010). Yeager et al. (2010) found that increased burden was associated with being an adult child and caregiver of the person with AD, as well as gender (female), and preexisting depressive symptoms (Yeager et al., 2010). Werner et al. (2012) conducted interviews of 185 adult child caregivers of older adults with AD, and found that behavioral problems, number of years caregiving, and caregiving stigmas of shame and decreased involvement were predictive of caregiver burden (Werner, Mittelman, Goldstein, & Heinik, 2012).

Conversely, structural equation model analysis highlighted positive aspects of caregiving in AD as opposed to the focus on burden. Relationships between religious coping, positive
aspects of caregiving, social support, burden, and depression were examined in one study utilizing a path analyses. The path analysis was a useful model because concepts are correlated both directly and indirectly. This was part of a multifaceted AD caregiving study with a sample of 648 respondents. The tested path model suggests that positive religious coping was positively associated with positive aspects of caregiving, and also indicates that those who have positive religious coping perceive a higher level of social support. Interestingly, negative religious coping was not related to positive aspects of caregiving nor social support. Further, the study confirms the importance of spirituality in AD (McGee, Myers, Carlson, Funai, & Barclay, 2013). Other research confirms burden was positively associated with depression (Heo, 2014).

Positive aspects of caregiving have been studied in the AD caregivers. One study found that 73% of caregivers of those with AD were able to identify at least one positive aspect of caregiving. These positive experiences, including companionship, a sense of reward, enjoyment, a sense of duty, providing quality of life, a sense of importance, and love for the care receiver, were associated with lower depression, burden, and better self-assessed health of the caregiver (Cohen, Colantonio, & Vernich, 2002). These studies highlight the importance of shifting focus to all aspects of caregiving, not just the negative.

Among this AD caregiving population, there are unmet service needs pertaining to activities of daily functioning (Li, Kyrouac, McManus, Cranston, & Hughes, 2012). Caregivers also request information and psychosocial support from healthcare providers, as well as financial support (Diehl-Schmid et al., 2013). Further needs for support for the caregiver include decision making around transition to residential care, driving ability, appropriate time for consulting other family members about decisions, initiation of hospice care, and supplemental feeding (Hoffman et al., 2013).
Cancer

Cancer is a chronic disease state in which those who are ill could potentially be very dependent on their informal caregivers. Caregivers of those with cancer have been shown to be at a significant risk for depression (Given et al., 2004; Lee, Lin, Chien, & Fang, 2015; Sklenarova et al., 2015; Stenberg, Cvancarova, Ekstedt, Olsson, & Ruland, 2014). One study examining a group of 164 spousal caregivers of patients with advanced lung cancer demonstrated social support was integral in ameliorating depression (Thielemann & Conner, 2009). Lower functional impairment of the care receiver was associated with increased psychological wellbeing of the caregiver. Satisfaction with social support also significantly affected psychological wellbeing of the caregiver (Goldstein, Concato, Fried, & Kasl, 2004; Ownsworth, 2010). Increased social support is a significant contributor to decreased caregiver burden in multiple studies (Burnette, Duci, & Dhembo, 2016; Chen et al., 2009; Kim, Duberstein, Sörensen, & Larson, 2005; Magliano, Fiorillo, De Rosa, Malangone, & Maj, 2005; Phillips, Gallagher, Hunt, Der, & Carroll, 2009). Other studies confirm presence of caregiver burden, distress, and anxiety in the context of caring for an individual with cancer (Fujinami et al., 2015; Kulkarni et al., 2014; Santin, Treanor, Mills, & Donnelly, 2014; Ugalde, Krishnasamy, & Schofield, 2014). Caregivers have also been shown to have more distress and anxiety than their care receivers (Sklenarova et al., 2015). Higher caregiver burden and stress were linked to decreased patient quality of life and higher depressed mood in the caregiver (O'Hara et al., 2010). In addition to depression, caregivers, especially adult children caregivers, reported a sense of abandonment (Given et al., 2004). Interestingly, this emotional burden did not seem to affect physical health despite the clear presence of burden in one study by Rha et al. (2015). In this study, emotional burden had no significant influence on physical activity, diet, smoking, and
alcohol consumption. These caregivers were actually more likely to adhere to cancer screening tests (Rha, Park, Song, Lee, & Lee, 2015). Despite this, caregivers continue to identify burden as their most important indicator of their quality of life, with open expression of feelings and household maintenance as important mediators in burden (Tamayo, Broxson, Munsell, & Cohen, 2010).

Informal caregivers of those with breast cancer are unique, as this population tends to be male and there is typically a spousal relationship. These caregivers had similar mental functioning and levels of depression as their care receivers, but these caregivers were markedly more anxious. Caregivers were also significantly more depressed in the transition to a terminal period as opposed to palliative. The majority of caregivers reported missing or having an impact on work, with prescription drugs being a major financial stressor. Burden was a predictor of both anxiety and depression (Grunfeld et al., 2004). Further, they experience fatigue and low quality of life (Gaston-Johansson, Lachica, Fall-Dickson, & Kennedy, 2004). Similarly, caregivers of those with recurrent breast cancer have similar emotional and well-being impairments as their care receiver, but report a worse relationship with the doctor, consisting of less confidence and less perceived availability to answer questions. Both patients’ and caregivers’ mental health scores were below the level reported for a sample of seriously ill patients, indicating presence of mental health dysfunction in this subset of caregivers (Northouse et al., 2002). Psychological impact does not lessen over time in this group (Khanjari, Langius-Eklöf, Oskouie, & Sundberg, 2014). In one study examining only husbands of those with breast cancer, they reported lower quality of life, higher extent to which emotions interfere with work and daily activities, and lower mental health as compared to control participants (Wagner, Bigatti, & Storniolo, 2006).

An important factor highlighted in this population of male informal caregivers is need to have a
partner who is more emotionally resourceful, as having less of this trait had a negative impact on men’s physical health (Kim et al., 2008).

Needs in this population caring for those with breast cancer include emotional support and a sense of security with a responsive health care team (Totman, Pistrang, Smith, Hennessey, & Martin, 2015), as well as instilling basic confidence in their ability to provide care (Beach & White, 2015). Communication with the healthcare team and increased education about medications and managing side effects were also identified as important needs to these caregivers (Tamayo et al., 2010). Caregivers also felt the need to be more involved in decision-making, have improved management of stress and more development of coping skills, as well as increased financial and psychological support (Santin et al., 2014).

**Dyspnea and Post-ICU**

With patients on prolonged mechanical ventilation, the majority of informal caregivers felt that employment was a major stressor. These caregivers also felt depression and interruption of lifestyle were burdens. Caregivers of patients in the post-intensive care unit phase are at greater risk for depression, anxiety, and post-traumatic stress disorder (Haines, Denehy, Skinner, Warrillow, & Berney, 2015; van Beusekom, Bakhshi-Raiez, de Keizer, Dongelmans, & van der Schaaf, 2015; Van Pelt et al., 2007), as well as clinically significant fatigue (Choi et al., 2014). Depressive symptoms were reported in caregivers of those who were intubated for more than 48 hours and these symptoms persisted at one year post-admission. Risk factors for caregiver lifestyle disruption include patient male gender, presence of functional dependency, patient education at least 12th grade, and patient tracheostomy (Haines et al., 2015). In patients with tracheostomies, caregivers of those requiring mechanical ventilation felt strain in the one-year follow-up from the original study, indicating that caregiver burden does not decrease over time.
They felt the need for additional teaching and felt that their lives were generally interrupted by caregiving (Ferrario, Zotti, Zaccaria, & Donner, 2001).

Need for social support, education, and assistance with activities of daily living are overarching themes in the literature involving caregiver burden. Risk factors for caregiver burden include being female, lower obtained level of education, living with the care receiver, higher number of caregiving hours, depression, social isolation, financial stress, and lack of ability to choose being a caregiver (Adelman, Tmanova, Delgado, Dion, & Lachs, 2014). Presence of burden in various disease states is the overarching theme within the review of the literature. There is a paucity of literature addressing positive aspects of caregiving beyond that of the AD population, creating a clear need for study of this topic within other disease states.

**Model Case**

PD, an 83-year-old woman, lives in a second-floor apartment with her husband, RD, who is also 83-years old. RD had a hip replacement five years ago and he has recently been diagnosed with Alzheimer’s disease, which has been a long process for PD. She has been managing his medication for the past two years, as he was becoming forgetful which she had previously attributed to older age before his diagnosis. His memory loss has most recently disrupted almost every aspect of their daily life and PD finds that RD has difficulty completing puzzles, his favorite activity, and that he cannot get to or from appointments without losing track of time or getting lost. He has also withdrawn from his family, refusing visits from their seven grandchildren.

PD is now responsible for managing the finances, shopping, making appointments, finding transportation for RD, and providing emotional support. She also is providing physical
support for RD as he has had a hip replacement and lives on the second floor. She finds ways to adapt and solve problems, even leaving directions for the microwave settings so RD is able to eat a snack if she happens to be away. PD loves and feels a sense of duty to RD after 60 years of marriage, and takes pride in her husband still living at home with her despite these difficulties. In this case study, PD’s experience with caregiving is clearly multidimensional. The experience has financial, social, emotional, and physical implications. The experience can be both negative and positive. It is important to not immediately label the experience as “burden,” as there are many positive aspects she is experiencing as well. The case contains all of the critical attributes of the caregiver experience, and provides examples of antecedents (RD requiring care) as well as consequences (PD’s burden as well as pride) (Walker & Avant, 2005).

**Conclusion**

Caregiver burden continues to have significant impact globally pertaining to cost, as well as individually with caregivers’ struggles with depression, anxiety, role strain, hours spent caregiving, and lack of social support. Despite this, caregivers are resilient and continue to perceive positive aspects to caregiving, although studies on positive reactions to caregiving are limited. Essentially, presence of these positive aspects is contrary to the conceptual meaning of burden, but they are inextricably linked in the caregiver’s experience. The caregiver’s experience is clearly a multidimensional concept that should be investigated without the bias of the lens of burden, essential in objective and impartial inquiry.
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Caregiver Reaction Assessment (CRA) for caregivers to persons with chronic physical

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Chapter 3: Measuring the Caregiver’s Experience

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Abstract
This article presents and reviews seven instruments used in the literature to quantify caregiver reactions. Instruments evaluated included the Bakas Caregiving Outcomes Scale (BCOS), the Caregiver Reaction Assessment (CRA), the Modified Caregiver Strain Index (MCSI), the Zarit Burden Interview (ZBI), the Perceived Caregiver Burden Scale (PCBS), the Multidimensional Caregiver Burden Inventory (MCBI), and the Screen for Caregiver Burden (SCB) (Bakas, Champion, Perkins, Farran, & Williams, 2006; Given et al., 1992; Novak & Guest, 1989; Stommel, Given, & Given, 1990; Thornton & Travis, 2003; Vitaliano, Russo, Young, Becker, & Maiuro, 1991; Zarit, Reever, & Bach-Peterson, 1980). Initial reliability and validity of each instrument is discussed, as well as a brief explanation of the overall tool and its use in the literature. Conceptual frameworks and use of instruments to measure positive reactions to caregiving are also investigated.

Keywords: psychometrics, caregiver reactions, empirical referents, burden
Measuring the Caregiver’s Experience: Reactions to Caregiving

The caregiving experience historically has been referred to as “burden”, “stress”, or “strain” in the literature. Such negatively connotative words would be a misnomer to many caregivers. The concept of positive caregiving reactions emerged as recently as the early 1990s (Ko, Yip, Liu, & Huang, 2008; Thornton & Travis, 2003) but caregiver burden remains the focus of measurement. Most commonly, caregiver burden is defined in the literature as “the extent to which caregivers perceive that caregiving has an adverse effect on their emotional, social, financial, physical, and spiritual functioning” (Zarit, Todd, & Zarit, 1986, pp. 260). In contrast, the positive aspects of caregiving, labeled as “caregiver reactions,” is defined as time devoted to caregiving, difficulty or ease of caregiving tasks, and overall perceived distress as well as positive outcomes such as high esteem and pride in caregiving (Chung, Pressler, Dunbar, Lennie, & Moser, 2010, pp. 412). The purpose of this chapter is to examine and compare the strengths and limitations of frequently-cited instruments used to measure the informal caregiving experience.

Review of Instruments

Bakas Caregiving Outcomes Scale

The Bakas Caregiving Outcomes Scale (BCOS) is fifteen-item scale that measures life changes resulting from providing care. Originally introduced by Bakas et al. in 2006 following a prior 10 item questionnaire, this tool was tested in a convenience sample of 147 family caregivers of stroke survivors, four months after the event. Nineteen percent of the stroke survivors were aphasic, and the majority of the caregivers were female (79%). The instrument is based on the Lazarus transaction-based approach to stress, where personality factors and caregiving tasks are mediated to produce caregiving distress or adaptive outcomes.
Questionnaires were administered by telephone or face-to-face interviews. Criterion-related validity was established using additional measures including the Stroke-Specific Quality of Life Scale Proxy Version (SSQOL- to determine level of impairment by caregivers), the Revised Life Orientation Test (to measure caregiver optimism), the Oberst Caregiving Burden Scale, the Appraisal of Caregiving Scale, the Patient Health Questionnaire Depression Scale, the Short Form-36 General Health Subscale (for caregiver perceptions of their own health), and finally the 15-item BCOS.

The BCOS uses a seven point Likert scale, with responses ranging from -3 (“[life has] changed for the worst”) to +3 (“[life has] changed for the best”). These scores translate to a 7-point scale for data analysis purposes. Higher scores indicate positive caregiver responses. Internal consistency reliability was demonstrated with a Cronbach’s alpha coefficient of 0.90. In the test retest reliability, in which a small sample of caregivers of aphasic stroke survivors took part, the authors obtained a Pearson rank correlation of 0.81 and an intraclass coefficient (ICC) of 0.66, indicating reliability.

Criterion related validity was established using a Pearson $r$ correlation between the BCOS and SF-36. Construct validity was established with a confirmatory factor analysis, which loaded a one factor solution, indicating unidimensionality. Forty-three percent of the variance was explained by the constructs in the conceptual model. The authors also performed a hierarchical multiple regression in which the BCOS was the dependent variable. Eleven independent variables were significantly related to the BCOS: SSQOL language, thinking, personality, self-care, survivor and caregiver gender, caregiver difficulty with tasks, depressive symptoms, appraisal of threat, appraisal of the benign, and appraisal of benefit (Bakas, Champion, Perkins, Farran, & Williams, 2006), indicating further construct validity.
The BCOS was used in one study on heart failure (HF), in which 100 family caregivers of HF patients were administered the BCOS in addition to several other tools to test the reliability and validity of a new 16-item Family Caregiver Quality of Life Scale (FAMQOL). A Pearson correlation of \( r = 0.73 \) between the BCOS and the FAMQOL provided evidence of criterion related validity for the BCOS. Limitations with the BCOS were weaknesses in detecting change and lack of spiritual wellbeing questions (Nauser, Bakas, & Welch, 2011). Spiritual wellbeing is important to address because it has been demonstrated to have a significant relationship with quality of life (Allahbakhshian, Jaffarpour, Parvizy, Haghani, 2010).

The BCOS also was used in a convenience sample of 63 family caregivers of patients with HF in a study by Pressler et al. (2009). The alpha coefficients reported in this study for the BCOS were 0.88, 0.72, and 0.82, at baseline, 4 months, and 8 months, respectively, providing evidence of internal consistency. The study found that caregivers with depressive symptoms were more likely to report their lives had changed negatively. At the 8-month testing, the perceptions of the caregiving experience did improve, but health-related quality of life was moderately poor over time (Pressler et al., 2009). Additional use of the BCOS in a sample with HF obtained a reliability coefficient of 0.88 (Bakas, Pressler, Johnson, Nauser, & Shaneyfelt, 2006).

Benefits of using the BCOS include sensitivity to appraisal of providing care and inclusion of a financial wellbeing question to assess financial burden. The BCOS is well suited as an assessment tool or outcome measure with its established reliability and validity. It has a higher alpha coefficient than its 10-question predecessor (\( \alpha = 0.85 \)), and is based on a well-established theoretical framework. There is also an item about the caregiver’s relationship with...
the stroke survivor, which potentially enables the inclusion of positive reactions to caregiving. The instrument requires 10-15 minutes to complete and is available for public use without cost.

Limitations include lack of generalizability due to a homogenous sample. The sample was mostly female, and only contained caregivers of stroke survivors, making findings only generalizable to this specific population. The two-week test retest only included caregivers of aphasic stroke survivors, possibly decreasing the reliability of this tool for non-aphasic populations. Although the authors describe the “potential” of the tool’s sensitivity to change, there is no actual evidence to support this (Bakas, Champion et al., 2006, pp. 8).

The Caregiver Reaction Assessment

The Caregiver Reaction Assessment (CRA) is an instrument that measures negative and positive reactions to caregiving. The multidimensional tool contains 21 items, with a 5-point Likert scale ranging from 1 (strongly agree) to 5 (strongly disagree). Developed in 1992 by Given et al., the original study utilized a convenience sample of 377 caregivers of elderly (>64 years old) adults in Michigan for the exploratory portion of the tool. The majority of the care receivers in the sample had physical impairments (71%), while others had Alzheimer’s disease (AD) or dementia (29%). The authors formulated 111 items with six dimensions in mind: the caregiver’s health, daily schedule, finances, perception of negative impact of caregiving, and how caregiving relates to one’s sense of self-worth. These items were reduced to 40, and tested with the above sample using an exploratory factor analysis (EFA). In the EFA, a component was retained if it explained greater than 5% of the variance.

The EFA produced five components, identified as caregiver esteem (7 items, $\alpha=0.90$), lack of family support (5 items, $\alpha=0.85$), impact on finances (3 items, $\alpha=0.81$), impact on
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schedule (5 items, $\alpha=0.82$), and impact on health (4 items, $\alpha=0.80$), reducing the tool to 24 items.

In the confirmatory phase, a new sample of 377 caregivers of those with AD and cancer was selected using the same criteria for a longitudinal analysis. This phase tested sensitivity of the factors to change in caregivers’ attitudes over time, with follow-up at six months and one year. All factors were shown to remain stable over time. Internal consistency reliability was demonstrated with alpha coefficients ranging from 0.80-0.90.

Criterion related validity was demonstrated with correlations between the ADL Dependency Scale and the Center for Epidemiological Studies Depression Scale (CES-D). Highest correlations were between: impact on schedule and CES-D/ADL ($\alpha=0.46$ and 0.52, respectively), and CES-D and impact on caregiver health ($\alpha=0.57$), with all components demonstrating statistically significant correlations with both the CES-D and ADL scale (Given et al., 1992).

The CRA has been used extensively in the cardiopulmonary population. In one study, the tool was administered to 18 caregivers of 20 HF patients requiring inotropic intravenous infusions and found to have a reliability coefficient ranging from 0.56 (caregiver health) to 0.84 (finances). The study found increased esteem and mental health of the caregiver increased quality of life in the care recipient. Caregivers were primarily spouses and felt positive about their role as caregiver, but the role had a negative impact on schedule, health, and finances. (Scott, 2000). In another sample of 50 caregivers and 41 HF patients, the CRA was administered via interview with participants. The alpha coefficient for the CRA in the sample was 0.85 overall, with the subscales as follows: esteem $\alpha=0.79$, family support $\alpha=0.88$, finances $\alpha=0.82$, schedule $\alpha=0.82$, and health $\alpha=0.81$, indicating internal consistency of the scale and its associated dimensions among HF patients (Saunders, 2008). Caregiver age, increased caregiving
hours, more caregiver health problems, caregiver depression, and increased patient comorbidities explained 51% of the variance in the CRA score. Additionally, in a sample of 29 patient-informal caregiver dyads awaiting pulmonary transplant, internal consistency reliability yielded an $\alpha$ of 0.90. Subscale coefficients ranged from $\alpha=0.67$ to $\alpha=0.90$. The caregivers felt privileged to care for the care recipient, but many (47%) also felt that caregiving has become central to their lives (Lefaiver, Keough, Letizia, & Lanuza, 2009).

Benefits of using the CRA include allowing for positive responses and a more comprehensive investigation into the caregiver’s experience. The ability to measure group differences as well as changes over time in caregivers is beneficial. The assessment is slightly longer than the BCOS, but measures five dimensions. Publicly available without cost, this scale is best suited for telephone or one-on-one interviews due to its need for personalization. Oral administration of the CRA is time intensive and may be viewed as a limitation. There was also no specification in the original validation study if the caregivers had medical training, which could confound results. Prior medical knowledge could potentially ease stress with caregivers, as there would be no learning curve or need for education by medical professionals. There was also very limited discussion of the confirmatory phase and capturing change over time by the authors.

**Modified Caregiver Strain Index**

The Modified Caregiver Strain Index (MCSI) is a 13-item tool introduced by Thornton and Travis in 2003. The multidimensional tool is scored from 0 to 2, with 0 being “no”, 1 being “sometimes”, and 2 being “yes”. The tool is a modification of the Caregiver Strain Index by Robinson (1983) and geared towards a geriatric population. It has an overall alpha coefficient of 0.86 and dichotomous answer options (Robinson, 1983). Thornton and Travis proposed the
Burden, Stress, and Strain Model, and the MCSI was developed from this framework (Thornton & Travis, 2003). The model proposes that care recipient’s mental/physical/global functioning, medication complexity, age of caregiver, age of care recipient, and length of time caregiving all contribute to caregiver burden. Medication administration challenges contribute to stress, which also contributes to the strain index of the caregiver.

For instrument testing, the authors recruit a sample of 158 family caregivers providing care for community-dwelling adults aged 53 years or older living, and requiring daily pharmacologic therapy. Internal reliability was established with an alpha coefficient of 0.90. Two-week test retest reliability for one-third of the sample (N=53) had an ICC of 0.88, again supporting reliability. Criterion-related validity was established with an exploration of the relationship between the MCSI and the original CSI, as well as the Family Caregiver Medication Administration Hassles Scale (FCMAH). With the CSI, the MSCI was significantly correlated at the p<0.01 level with mental functioning ($r=0.34$), physical functioning ($r=0.27$), and global functioning ($r=0.32$). No significant relationship was established with the FCMAH because of differences in measurement (Thornton & Travis, 2003).

MCSI usage is sparse in the literature. The MCSI was given to a sample of 61 family caregivers in Singapore to assess relationship between caregivers’ stress, gender, and the functional status of the care recipient. The MCSI had an alpha coefficient of 0.82 in this sample. The study established a direct relationship between levels of stress and physical dependency of the patient. Females and those taking care of older adults with dementia, Parkinson’s disease, and hypertension experienced significantly greater stress (Mehta, 2005).

The MCSI has many benefits. It can be administered quickly in interview situations or by survey. The tool is readily available without cost. It is a newer instrument with an increased
reliability coefficient compared to the older CSI. Unfortunately, the tool has not been used extensively so there is limited construct validity in other populations (e.g. caregivers of those with chronic health conditions), limiting generalizability. In addition, the MCSI uses the caregiver’s self-report, potentially affecting reliability and validity. Lack of published literature utilizing this instrument is a weakness, as validity and reliability has not been established in various care receiver disease states. The focus on objective burden also could potentially limit the utility of this tool. There are no subjective items included in the tool. The focus on objective caregiver burden could be a strength or a weakness depending on the outcome variable being measured.

**Zarit Burden Interview**

The Zarit Burden Interview (ZBI), introduced in 1980, is widely used for assessing burden in caregivers of those with dementia. It is the oldest caregiver burden tool. It consists of 29 items, although a 22, 12, and 4 item tool exist. Answers are arranged using a five point Likert scale ranging from 0 (“never”) to 4 (“nearly always”). The tool has a two-factor structure consisting of role strain and personal strain. The authors do not mention the use of a theoretical framework.

The ZBI was evaluated with a sample of 29 caregivers of persons with AD. The survey results for the sample had an alpha coefficient of 0.85, indicating reliability. The ZBI has consistently had an alpha coefficient above 0.80 in AD studies (Harkness & Tranmer, 2007). Construct validity was established correlating survey results with the Kahn and Jacob’s mental status questionnaire, as well as functional ability, frequency of memory/behavioral problems,
duration of illness, personal activities of daily living (PADLs), instrumental activities of daily living (IADLs), and frequency of family visits (Zarit, Reever, & Bach-Peterson, 1980).

The ZBI was utilized in two prominent studies on patients with HF. In one study by Trivedi et al. (2011), relationship between spousal well-being and care receiver’s health was examined in a sample of 23 veterans with heart failure (HF). Using the sample of caregivers (100% female), the ZBI had an alpha coefficient of 0.88. Mean caregiver burden score was 22.3, in the “high” range. Fifteen spouses (75%) reported ZBI scores of >17, indicating extreme burden. Burden was related to spousal depressive symptoms (r = 0.48), and ratings of patients’ functional limitations (r = -0.50) at the p<0.05 level (Trivedi, Piette, Fihn, & Edelman, 2012).

Chung et al. (2010) examined differences between caregivers with and without depressive symptoms using a sample of 109 caregivers and patients with HF (mean age=57 years old; 79% spousal caregivers, 75% female). The alpha coefficient for this sample was 0.93. The mean ZBI score was 15.9 (SD= 13.1), and 33% of caregivers had severe distress related to caregiver burden. Caregivers’ own functional disabilities, perceived control, and caregiver burden explained 45% of the variance in caregivers’ depressive symptoms (Chung, Pressler, Dunbar, Lennie, & Moser, 2010).

Some benefits to using the ZBI are that it is free to use, and it is frequently present in the caregiving literature. The ZBI also has been translated and validated in 18 different languages (Harkness & Tranmer, 2007). It is also available in varying lengths. Availability of a shorter instrument could benefit a smaller sample size for an adequate power analysis. Its brevity can also help in a time constrained situation. The ZBI also can be given in an oral interview as well as in a survey. Unfortunately, this instrument does not include positive reactions to caregiving, and it has limited use in populations other than caregivers of those with AD. Additionally, a
sample of only low-severity AD patients was included in the original development of the instrument. As a result, the tool may not accurately capture severe burden, raising questions about its reliability and validity across the full spectrum of caregiver burden. In addition to this, there was no mention of a conceptual framework for the ZBI.

**The Perceived Caregiver Burden Scale**

The Perceived Caregiver Burden Scale (PCBS) was initially published in 1990 by Stommel, Given, and Given, and later revised into a shorter instrument in 1999 by Gupta. In the original instrument, all but five of the 31 items rated are reverse coded using a Likert scale of 1, indicating low burden, to 4, indicating high levels of burden. Initially, the PCBS had five subscales with alpha reliability coefficients ranging from 0.72 to 0.92. These subscales include: impact on finances, impact on work schedule, feelings of abandonment by extended family, impact on health of the caregiver, and sense of entrapment (Stommel, Given, & Given, 1990). The PCBS was later revised by Gupta (1999), utilizing a sample of Pakistani caregivers (N=150). The confirmatory factory analysis suggested a poor fit with the 31-item, 5-factor structure initially reported by Stommel et al. (1990). Based on inter-item correlations, several items were eliminated, resulting in a 12-item scale and three constructs. The constructs are as follows: impact on finances ($\alpha=0.89$), impact on work schedule ($\alpha=0.90$), and sense of entrapment ($\alpha=0.96$). There is no mention of a theoretical framework. Gupta validated the original instrument as well as the 12-item instrument in Hindi (Gupta, Gupta, & Gupta, 1999; Stommel, Given, Given, & Gupta, 1999).

The PCBS was used several times, most notably by Gupta to reaffirm reliability and validity, and in individuals who abuse drugs in Vietnam. One study with a sample size of 259
caregivers of older adults in India found three subscales: impact on finances ($\alpha=0.77$), abandonment by extended family ($\alpha=0.81$), and sense of entrapment ($\alpha=0.82$), along with concurrent validity with the variables of role overload, role conflict, age of caregiver, as well as the Center for Epidemiological Studies Depression Scale (CES-D) (Gupta, 2007). Lee et al. (2015) also found significant caregiver burden, although HIV positive versus HIV negative status of the individual was not significant ($p=0.2105$). Additionally, among families who were retested at three months, and then again at six months, burden levels were fairly consistent for the family members of HIV negative drug abusers. However, burden increased among the family members of those drug abusers who are HIV positive (Lee, Li, Lin, & Tuan, 2015).

The benefits of the PCBS include free cost, and flexibility with administration. The instrument was translated and validated in Hindi but no other languages. It is also multidimensional and the short form increases ease of data entry, as reverse coded items are removed. Internal consistency reliability has been established. Weaknesses include the length of the long form instrument, the nature of self-report, lack of theoretical framework, and initial negatively coded items in the longer Stommel et al. (1990) version of the instrument. Gupta later removed these items based on low factor loading, less than 0.20, likely indicating participants’ difficulty with understanding the reverse coded questions, as they were negatively worded (Gupta, 1999). Additionally, there is no inclusion of positive reactions to caregiving.

The Multidimensional Caregiver Burden Inventory

The Multidimensional Caregiver Burden Inventory (MCBI), published originally in 1989, examines caregiver burden in 107 caregivers of confused or disoriented older people. The majority of the sample was female (73.8%), with half of the sample caring for the older adult in
the home, and half in an institution. Unlike prior studies with an adult child caregiver or an undefined caregiver-care receiver relationship, spouses comprised 44.9% of the caregivers. The MCBI consists of 24 items; all presented using a Likert scale of 0-4, with items ranging from 0 (“not at all descriptive”) to 4 (“very descriptive”). Upon factor analysis, five factors loaded, which accounted for 66% of the variance. The factors are described as follows: time-dependence burden ($\alpha=0.85$), developmental burden ($\alpha=0.85$; described as caregivers’ perceived development being less than their peers), physical burden ($\alpha=0.86$), social burden ($\alpha=0.73$), and emotional burden ($\alpha=0.77$) (Novak & Guest, 1989).

The MCBI was used in a sample of low-income women transitioning from welfare to employment (N=31) who were providing informal caregiving to another person in their family or community. Burden scores using the MCBI, which can range from 0 to 72, was “moderate” with a mean of 30.4 (range= 6-68). There was no significant relationship between job scheduling flexibility and caregiving burden ($r=0.326$, $p=0.147$), although this was very much subject to type 2 error as the sample size was small. Highest amount of caregiving burden fell into the time-dependence dimension of the MCBI. The authors were unable to determine a factor structure for the MCBI due to too few participants identifying as informal caregivers (Kneipp, Castleman, & Gailor, 2004).

Benefits of using the MCBI include free cost, multidimensionality, validation in Chinese (Chou, Jiann-Chyun, & Chu, 2002), and internal consistency reliability. Possible shortcomings of the instrument include self-report of participants, no established concurrent reliability with other existing instruments, and lack of recent frequent use in the literature.

Screen for Caregiver Burden
The Screen for Caregiver Burden (SCB) was developed by Vitaliano et al. (1996) using various samples of spousal caregivers of those with Alzheimer’s disease. In phase I of instrument development, 68 dyads were interviewed about subjective and objective burden and the tool was developed from these caregiving concerns. Concurrent reliability was established with the Beck Depression Inventory, as well as the Sickness Impact Profile/the functional status of the spouse with Alzheimer’s disease. In phase II of instrument development, 79 dyads were recruited for a longitudinal investigation. Phase III consisted administering the SCB to 24 dyads, with a control group of spouses in non-Alzheimer’s disease group (N=20) for demographics, health, and psychological distress within the group to establish criterion validity. Total sample size in all phases was 191 (Vitaliano, Russo, Young, Becker, & Maiuro, 1991).

The SCB consists of 25 items, scored on a 5-point Likert scale. The instrument has two dimensions as defined by the authors; subjective and objective burden. There was no mention of a confirmatory factor analysis. Alpha coefficients were sufficient for the two dimensions (subjective $\alpha=0.84$, and objective $\alpha=0.89$). Over time, test-retest reliabilities were $\alpha=0.70$ and $\alpha=0.64$ respectively. There was no mention of a theoretical framework.

One study by Davis et al. (2006) with a sample of 131 caregivers (mean age=63.7 years; care recipient mean age= 75.5 years) found an objective burden $\alpha=0.82$, and a subjective burden $\alpha=0.91$. Average objective burden was measured at 8.4, and subjective burden was measured at 14.8, indicating moderate burden (Davis, Weaver, & Habermann, 2006).

Benefits of the SCB include rigorous instrument development with a large sample size and multiple phases, concurrent reliability, internal consistency reliability, criterion validity, construct validity, and ease of use. Unfortunately, the SCB has not been utilized in recent literature, and the subjective and objective domains were not confirmed by a factor analysis. The
focus of this instrument is burden, so there is no appraisal of positive reactions in caregiving. There is also no mention of a theoretical framework. Additionally, the SCB has only been validated in English and is longer (25 items) in comparison to other instruments.

**Conclusion**

Analyzing caregiver reactions remains challenging, even with the availability of valuable instruments to assist in quantifying an otherwise abstract concept. Each instrument has valuable input. The ZBI has extensive presence in the literature since the 1980s. The MCSI could be utilized as an objective measure of strain and stability over time. The BCOS is beneficial for its inclusion of all life changes for the caregiver, and the PCBS for its inclusion of abandonment by extended family members and validated translations. The MCBI could be used for its five domains and inclusion of development of the caregiver, and the SCB for its rigorous development and validation. Notably, the CRA includes positive reactions to caregiving. The best instrument is dependent on its indication for use, as all the reviewed instruments demonstrated reliability and validity. As can be noted in Appendix A, each instrument measures a slightly different domain of the caregiver experience, making the finer nuances of the phenomenon in question important in selecting an instrument. Instruments, such as the selected ones reviewed, assist in more easily measuring a concept such as caregiver reactions. Concrete, objective data can be collected from caregivers with these instruments to better demonstrate the caregivers’ needs. This in turn enables study on a larger scale involving interventions, an integral part of scholarly inquiry.
References


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## Appendix A

**Table 1**

*Instruments to Measure Caregiver Burden*

<table>
<thead>
<tr>
<th>Instrument</th>
<th>Original Population Used</th>
<th>Domain</th>
<th>Description</th>
<th>Validity</th>
<th>Reliability</th>
</tr>
</thead>
<tbody>
<tr>
<td>BCOS (2006)</td>
<td>Caregiver of stroke survivors</td>
<td>Appraisal</td>
<td>15 items, modification of 10-item scale. Likert scale of -3 (“changed for the worst”) to +3 (“changed for the best”).</td>
<td>CFA= unidimensional Criterion Construct</td>
<td>α= 0.90 2-week test retest= 0.66</td>
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<tr>
<td>CRA (1992)</td>
<td>Caregivers of patients with physical limitations, Alzheimer’s disease, dementia, cancer</td>
<td>Reaction</td>
<td>21 items, 5 point Likert scale of 1 (“strongly agree”) to 5 (“strongly disagree”). Positive responses.</td>
<td>EFA= 5-factor model Construct Content</td>
<td>Factor structure stable 6mon-1yr Internal consistency: Schedule= 0.82 Financial= 0.81 Support= 0.85 Health= 0.80 Esteem= 0.90</td>
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<tr>
<td>MCSI (2003)</td>
<td>Caregivers of &gt;53 year old adults living in community based settings, taking medications daily</td>
<td>Strain</td>
<td>13 items, scored 2 points= “yes”, 1 point= “sometimes”, and 0 points= “no”.</td>
<td>Construct</td>
<td>α= 0.90 Two week test retest= 0.88</td>
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<tr>
<td>Test</td>
<td>Group</td>
<td>Content Constructs</td>
<td>Rating</td>
<td>Notes</td>
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<td>ZBI (1980)</td>
<td>Caregivers of cognitively impaired elders</td>
<td>Burden</td>
<td>Interview, original=29 items but shortened to 22, 12, and 4 items. Two factor structure= personal strain and role strain. Likert scale from 0 (“never”) - 4 (“nearly always”). Translated into 18 languages. Used very frequently in caregiving literature.</td>
<td>Content Construct</td>
<td>α=0.85, consistently α&gt;0.80 in literature related to dementia</td>
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<tr>
<td>PBCS (1990)</td>
<td>Caregivers of the elderly</td>
<td>Burden</td>
<td>31 items on original instrument (some reverse coded), shortened to 12 items. Revised instrument with three factors: impact on finance, impact on work schedule, and sense of entrapment. Likert scale from 1 (“low/no burden”) to 4 (“high levels of burden”). Reverse coded items removed on short form.</td>
<td>CFA= 3-factor model (1999) Concurrent (correlated with CES-D)</td>
<td>α of revised factor loadings= 0.89, 0.90, 0.96.</td>
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<tr>
<td>Study</td>
<td>Description</td>
<td>Burden</td>
<td>CFA Model</td>
<td>Validation</td>
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<tr>
<td>MCBI (1980)</td>
<td>Caregivers of confused or disoriented older people</td>
<td>24 items, Likert scale of 0 (“not at all descriptive”) to 4 (“very descriptive”). Loaded five factors on factor analysis: time-dependence burden ($\alpha=0.85$), developmental burden ($\alpha=0.85$), physical burden ($\alpha=0.86$), social burden ($\alpha=0.73$), and emotional burden ($\alpha=0.77$). Validated in Chinese.</td>
<td>5-factor model</td>
<td>No overall $\alpha$ reported. $\alpha$ of factors= 0.85, 0.85, 0.86, 0.73, and 0.77.</td>
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<td>SCB (1991)</td>
<td>Spousal caregivers of those with Alzheimer’s disease</td>
<td>25 items, 5-point Likert scale, two dimension. Subjective dimension $\alpha=0.84$, and objective dimension $\alpha=0.89$</td>
<td>Construct Criterion Validity</td>
<td>Subjective dimension $\alpha=0.84$, and objective dimension $\alpha=0.89$</td>
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Chapter 4: Caregiver Reactions in Pulmonary Hypertension

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Abstract

This phenomenological, qualitative study investigates the informal caregivers’ reactions to caring for individuals with pulmonary hypertension (PH). There is a paucity of research involving PH informal caregivers. While the disease itself has been shown to be burdensome, caregivers are not addressed (Flattery, Pinson, Savage, & Salyer, 2005). The sample includes caregivers of those with New York Heart Association (NYHA) functional class II-IV PH. Participants were recruited from a Boston PH support group, and the Pulmonary Hypertension Association (PHA) online support forum. The majority of care receivers were World Health Organization (WHO) group I (57.1%), and were on continuous oxygen (57.1%). The average age of the caregivers (n=7) was 63.1, with the majority being female (71.4%). Methods utilized include open-ended semi-structured interview questions. Analysis was completed using Moustakas’ modification of the Stevick-Colaizzi-Keen method of phenomenological investigation, with two-cycle coding. Data were then organized and clustered into patterns using a word cloud followed by Pattern coding. Findings included six overarching themes. Participants described the initial phase after diagnosis as overwhelming, with a long time between first symptom and diagnosis. There was a period of having to become clinically literate and learn to deal with medications and side effects. Participants described entering a new normal to cope with PH, but there was still a certain amount of stress that came with caregiving. Many participants were dual caregivers, adding to this challenge. Further, all participants encouraged seeking out support and means of self-care, and described an improvement in their relationship with their care receiver.

Keywords: pulmonary hypertension, informal caregivers, caregiver reactions
Caregiver Reactions in Pulmonary Hypertension

Pulmonary hypertension (PH) is a chronic, progressive disease that results in a multitude of symptoms including dyspnea, pain, anxiety, cyanosis, dizziness, syncope, and edema (Matura & Carroll, 2010). The annual number of hospitalizations for PH has tripled among Medicare recipients, from 55,516 in 1990 to 187,205 in 2002 (Hyduk et al., 2005). Death rates among men and women are increasing, from 6,570 in 2006 to 8,261 in 2010 in men, and from 10,644 in 2006 to 13,031 in 2010 in women. For those ages 85 years and older, hospitalization rates have doubled, and death rates are rapidly accelerating (George, Schieb, Ayala, Talwalkar, & Levant, 2014). All age groups are affected, especially in those aged 65 years and older. Evidence suggests a global PH prevalence of up to 10% in this age range (Hoeper et al., 2016). Prior to 1995, most patients with PH had a prognosis of less than three years after initial diagnosis (Barst, 2008). While highly variable with etiology of PH, prognosis has certainly improved today, but much of the life-sustaining therapy is costly and labor-intensive. Patients often require oxygen and other extensive therapies, including expensive, centrally administered medications (Holcomb, 2004; Steinbis, 2008). Home care and informal caregiving is arduous with these treatments. Despite the severity and increasing incidence of the disease, there is a paucity of literature regarding concepts of caregiver reactions within the context of PH. This qualitative study will describe reactions of caregivers ministering to adults with PH.

Background

Pulmonary Hypertension

Pulmonary hypertension was first recognized by the World Health Organization (WHO) in 1973 following an outbreak of PH related to the use of aminorex fumarate, an appetite
suppressant. The WHO has held five world symposiums on PH, with the latest in 2013 (Galiè et al., 2013; Fishman, 2004; Hoeper, 2009). The disease stems from a multitude of etiologies that the WHO has categorized into five groups. Etiologies vary from idiopathic (WHO group I), left heart disease related PH (WHO group II), lung disease associated PH (WHO group III), chronic thrombotic disease related PH (WHO group IV), and miscellaneous etiologies (WHO group V) (Ross, 2007; Simonneau et al., 2013). Appendix A contains a complete list of the WHO classifications of etiologies.

Epidemiology and prognosis of PH is difficult to track due to variance in etiology and severity among patients. Diagnosis is frequently elusive, as PH often presents with nonspecific symptoms, and with concurrent underlying disease (Yates & Saunders, 2008). PH diagnosis is typically made with a right heart catheterization and pulmonary artery pressures are measured at greater than 25 mm Hg at rest (Hoeper, 2009; Lourenço, Fontoura, Henriques-Coelho, & Leite-Moreira, 2012; Steinbis, 2008). Average time from initial symptoms to diagnosis is two to three years (Brown et al., 2011; Traiger, 2007) due to multiple differential diagnoses. Because PH can frequently be misdiagnosed as a cardiac or pulmonary problem, diagnosis confirmation often occurs following permanent damage to the pulmonary vascular bed (Tartavoulle, 2011). Deaths from PH increased from 10,922 in 1980 to 15,668 in 2002, with a greater increase observed in women. Death rate for PH as any contributing cause of death was 5.5 per 100,000 in 2001 and 6.5 per 100,000 in 2010 (George et al., 2014). Between 2000 and 2002, 807,000 patients were hospitalized with PH as one of their diagnoses; 61% were women and 66% were aged 65 or older (Hyduk et al., 2005). Global prevalence is estimated at 10% of those ages 65 and older with WHO group I PH (Hoeper et al., 2016). The prevalence and incidence vary widely because PH
frequently presents with many comorbid conditions, although the PH may not necessarily be independent of the concurrent disease.

Following diagnosis, there is wide variation among clinicians in determining WHO group and functional class (Steinbis, 2008; Taichman et al., 2005), making it more difficult to track specific statistics. Prognosis is often difficult to establish and has been cited as poor in cases of WHO group I (idiopathic) PH. The mean age at diagnosis for this group is 45 years, and 79% of those who exhibit the disease are female (Frost et al., 2010). The average life expectancy of WHO group I PH patients without treatment is frequently cited as about three years from the time of diagnosis (Adiutori, 2000; D’Alonzo et al., 1991; Lourenço et al., 2012; Yates & Saunders, 2008). Underlying or comorbid disease can create difficulty in estimating the outcome of the isolated PH (Yates & Saunders, 2008), resulting in no clearly established prognosis or age of onset in the other WHO etiology groups.

While there is no cure, symptoms are managed with use of diuretics, digoxin, calcium channel antagonists, oxygen therapy, and anticoagulants (Hoeper, McLaughlin, Al Dalaan, Satoh, & Galiè, 2016; Sastry, 2006; Steinbis, 2008). Treatment for PH can consist of endothelin receptor antagonists (bosentan and ambrisentan), phosphodiesterase-5 inhibitors (sildenafil and tadalafil), as well as prostacyclins (epoprostenol, treprostinil, and iloprost) (Pohar, Clark, & Spry, 2009; Tartavoulle, 2011). Rarely, certain calcium channel blockers are used (Hopkins, Rubin, Mandel, & Finlay, 2017). A more recent medication, riociguat, a soluble guanylate cyclase stimulator, was approved for use in PH in October of 2013 (Frey, Mück, Unger, Artmeier-Brandt, Weimann, & Wensing, 2008; Hoeper, McLaughlin, Al Dalaan, Satoh, & Galiè, 2016; Hopkins et al., 2017). The prostacyclin drug family is essential to New York Heart Association (NYHA) functional class III-IV PH patients, who are the most severely functionally
impaired, as they can be used intravenously as potent vasodilators in the case of failure with oral therapy and the treatment is among the most dangerous. (Oudiz et al., 2004; Smyth, Grosser, Wang, Yu, & Fitzgerald, 2009; Tartavoulle, 2011). Surgical options for PH patients include lung transplantation and pulmonary thromboendarterectomy when indicated (Haythe, 2012; Tartavoulle, 2011).

The pharmacological treatments for PH, while advanced, can be dangerous and expensive. Costs for intravenous (IV) medications ranged from an estimated $36,000 to $93,000 annually for the drugs alone, not including refrigeration and administration costs (Widemar, 2005). Multiple side effects are also problematic. The prostacyclins are most likely to pose a problem, with side effects such as systemic hypotension, nausea, vomiting, headache, edema, pruritus, vasodilation, jaw pain, irritation at infusion site, and musculoskeletal pain. Serious adverse effects include sepsis from central line infection, rebound pulmonary hypertension from acute withdrawal, and increased risk of hemorrhage (GlaxoSmithKline, 2016; United Therapeutics, 2014). These symptoms and treatments certainly comprise a large amount of the PH patient and caregiver experience, and are what differentiates PH from left-sided heart failure. Maintenance of a central line, a potent vasodilator, and the potential for acute respiratory distress and/or sepsis challenge both care receiver and caregiver.

**PH: The Patient Experience**

Although the phenomenon of interest in the proposed study is the experience of caregivers, it is important to first understand the PH patient experience, as the two concepts are inextricably linked. Nursing research on the PH experience is sparse, as many studies focus on pharmacological or pathophysiological aspects of the disease. Currently, nurses are just
beginning to investigate the patient experience of PH, with only one randomized controlled trial
to date (Taichman et al., 2005). Many studies are in the descriptive phase, with a predominantly
qualitative or correlational design (Flattery, Pinson, Savage, & Salyer, 2005; McDonough,
Matura, & Carroll, 2011). The majority of the studies describe psychiatric comorbidities, and
many use quality of life or six-minute walk test (6MWT) scores as outcome measures (Taichman
et al, 2005).

Only two studies to date attempt to describe the whole PH patient experience. The
authors in one study describe the lived experience of eleven patients with PH, in which two
overarching themes were “coping with uncertainty” and “life with treatment.” Information
seeking, especially using the internet, is a critical step when first diagnosed. Other common
themes include fear of the unknown and coping (Flattery et al., 2005). The other study examining
the PH patient experience among WHO group I PH patients used qualitative descriptive
methodology. The authors conducted semi-structured interviews with 10 patients with PH and
uncovered two overarching themes: holding back (fear, anticipating that symptoms will worsen,
and side effects of treatment) and redefining life (uncertainty, restrictions on daily activities, and
coping) (McDonough, Matura, & Carroll, 2011). These findings were similar to the work by
Flattery et al. (2005) and underscore the need for self-management interventions.

Narrative analysis of creative writing regarding the experience of PH was investigated by
Goddard et al. (2017). Detailed unstructured narratives initiated from open-ended writing
prompts aimed at building a “micro-story” about the patient’s illness were collected from twelve
patients attending a pulmonary vascular clinic in the United Kingdom. These narratives were
typically between 200-300 words long and examined the experiences of diagnosis. The
narratives also explored specialist services, symptom severity, family impact, and coping skills.
Overall results showed that PH had a profound effect on life, with ongoing and unpredictable needs requiring adaptation. The study highlighted the need for a more holistic form of treatment in PH, as diagnosis and continued treatment is life-altering (Goddard, Armstrong, Kiely, Elliot, Charalampopolous, Condliffe, Stone, & Sabroe, 2017).

The presence of psychiatric disorders in the PH patient is another important theme in the PH literature. Descriptive in nature, these studies differ from previous work because they focus solely on the mental health component of living with PH. In one study, 35% of individuals with PH suffer from concurrent psychiatric illness, with the most common being major depressive disorder (15.9%) followed by panic disorders (10.4%). Development of psychiatric illness greatly increases as functional limitations from PH increase, suggesting a correlation between functional status and psychological distress (Löwe et al., 2004). White et al. (2006) found cognitive sequelae occurred in 58% of the sample of 46 WHO group I PH patients. Twenty-six percent of this sample exhibited moderate to severe depression, and 19% had moderate to severe anxiety (White, Hopkins, Glissmeyer, Kitterman, & Elliott, 2006). Presence of depression in another study suggests a correlation with functional status (McCollister et al., 2010). A much older letter to the editor from 1986 found that three patients (without PH) who were started on a prostacyclin infusion developed moderate to severe depression, with immediate relief from stopping the infusion (Ansell, Belch, & Forbes, 1986). If this depression was prostacyclin-induced, a correlation between this drug class and depression could be problematic for the PH patient. Those with PH have a higher incidence of depression than the general population, with an estimated 26.2% of Americans ages 18 and older suffer from mental illness in any given year (Kessler, Chiu, Demler, Merikangas, & Walters, 2005). Löwe et al. (2004) reports an estimated 35% of PH patients suffer from psychiatric illness (Löwe et al., 2004).
Quality of life and the 6MWT in the PH patient were outcome measures frequently referenced. Taichman et al. (2005) found that in a sample of 155 individuals, those with WHO group I PH had the best quality of life, while those with PH related to systemic sclerosis had the worst health-related quality of life. Better 6MWT times also correlated with increased quality of life scores. Interestingly, hemodynamics did not correlate with quality of life scores, and no differences were found in quality of life among those taking oral calcium channel antagonists, oral bosentan, and IV epoprostenol (Taichman et al., 2005). Mereles et al. (2006) conducted the only non-pharmacological interventional study examining the effects of an exercise program on quality of life of PH patients. The program consisted of progressive bicycle and weight training in addition to sixty minutes of walking and respiratory exercises five days per week. The training regimen was shown to be beneficial in increasing self-reported quality of life as well as improving NYHA functional class (Mereles et al., 2006).

**The Caregiver’s Experience in Heart Failure**

The reaction of the caregiver is the focus of this study. The term “reaction” is used because it carries no negative or positive connotations, and generally is more active diction in comparison to “experience” or “burden.” The phenomenon of focus is the specific response to stimuli, in this case, a care receiver with PH, and specific actions subsequently taken for the caregiver to adapt. The overwhelming majority of the literature regarding caregivers focuses on burden as a variable instead of more neutral reactions. Caregiver experience literature review draws from informal caregiver research in other disease states due to the overall lack of PH-specific caregiver research. Investigation into caregiving for other disease states provides possible frameworks for understanding the experience of the PH caregiver.
There is one descriptive study that focused solely on positive aspects of caregiving by Cohen et al. (2002). The study found a correlation between identification of positive aspects in illness and lower depression scores, lower burden scores, and better self-assessed health. Some positive aspects of caregiving for the study’s sample of older adults with dementia included: companionship (22.5%), a sense of fulfillment (21.8%), enjoyment (12.8%), obligation (10.4%), and demonstration of love (5.5%) (Cohen, Colantonio, & Vernich, 2002).

Similar to the acute care needs of the PH population, a study by Gysels and Higginson (2009) examined caregivers of those with heart failure, COPD, motor neuron disease, and/or cancer. The study investigated how care is managed for an individual who suffers from breathlessness at home and had sample of 15 caregivers. Using a grounded theory approach, the most important finding was that caregivers were not very well equipped to deal with severe shortness of breath in the home setting. Negative aspects of caregiving included uncertainty, the caregiver’s own health problems, shock at diagnosis, perceived judgments of healthcare professionals, person (care receiver) loss, and acute exacerbations in the care receiver (Gysels & Higginson, 2009, pp. 3-4). Themes that contributed to successful care included acceptance, self-care, support, shared responsibility, and continuing with daily life (Gysels & Higginson, 2009).

Ågren et al. (2010) utilized the Caregiver Burden Scale in a study on partners of heart failure (HF) patients. The sample included 135 partners with a median age of 69 years (75% of whom were females). They found that 30% of the caregivers perceived burden as “medium,” and 2% reported burden as “high,” significant as almost one-third of the sample even perceived burden in the NYHA functional class I-III patient. NYHA class IV caregivers did not partake in the study, so the participants involved did not have many functional limitations. The most difficult area for caregivers was maintaining their own freedom and social interaction, as well as
having some amount of control over the cardiovascular care of their partner (Ågren, Evangelista, & Strömberg, 2010). Additional literature on this topic indicates that less family support correlates with higher HF patient functional impairment, as well as greater disruption to caregivers’ schedules. The caregiver’s health correlated with recent hospitalizations of the HF patient, as well as perceived control and social support (Boyoung, Fleischmann, Howie-Esquivel, Stotts, & Dracup, 2011).

The Caregiver’s Experience in Other Disease States

The prior studies are salient because of their relevance to symptoms and pathophysiology of disease in PH. However, the possibility of acute exacerbation is far greater in the PH population. This potentially may have important implications. Gysles and Higginson (2009) found that caregivers did not deal well with severe shortness of breath in the home in a qualitative study of caregivers caring for a person with advanced illness in the home. Dyspnea is difficult to handle for the lay caregiver, and that this could potentially create burden or stress for the caregiver. Social support was imperative to this population of caregivers, and they felt unprepared for an acute exacerbation. There has been no application of preparedness for dyspnea or an acute exacerbation to PH patients or caregivers. There was also an incorporation of positive influences to caregiving, including acceptance, self-care, availability of support, and feeling that caring is a shared responsibility (Gysles & Higginson, 2009).

A vast amount of the literature on caregivers relates to a cancer diagnosis for the care receiver. Caregivers within this population have been shown to be at a significant risk for depression. In one study examining a group of 164 spousal caregivers of patients with advanced lung cancer, social support was shown to reconcile the relationship between burden and depression (Thielemann & Conner, 2009), underscoring the importance of family and social
networks. With caregivers of those diagnosed with both brain tumors and other cancers, less functional impairment of the patient was associated with increased psychological wellbeing of the caregiver. Satisfaction with social support also significantly affected psychological wellbeing of the caregiver (Ownsworth, 2010). Multiple other studies cite the importance of social support in this caregiving population (Chen et al., 2009; Magliano, Fiorillo, De Rosa, Malangone, & Maj, 2005; Marsland et al., 2013; Phillips, Gallagher, Hunt, Der, & Carroll, 2009; Shieh, Tung, & Liang, 2012).

Caregivers of those with breast cancer are an important population because the patients are demographically similar to PH patients (i.e., mostly female) and treatment can be demanding and complex. In multiple studies with a male primary caregiver for a breast cancer patient, increased burden and decreased quality of life were reported (Grunfeld et al., 2004; Mancini et al., 2011; Wagner, Bigatti, & Storniolo, 2006; Wagner, Das, Bigatti, & Storniolo, 2011). Depression (Grunfeld et al., 2004; Kozachik et al., 2001) and domestic role strain (Wagner et al., 2011) are additional struggles specific to this population. Drug and alcohol use can also be a problematic means of coping for this primarily male caregiving population (Kershaw, Northouse, Kritpracha, Schafenacker, & Mood, 2004).

Palliative care is an important field when examining PH, as symptom control and avoiding exacerbation, as well as relieving pain or dyspnea, are central goals. One study involving informal caregivers of those on inpatient palliative care found five overall unmet needs of caregivers: 1) sensitive, effective communication about advanced illness; 2) timely access to coordinated medical care; 3) respect for and honoring care decisions; 4) psychological, social, and spiritual needs; and 5) caregiver support. The study was a video-taped, longitudinal,
ethnographic study utilizing semi-structured interviews. One of the twelve participants had a diagnosis of PH (Tallman, Greenwald, Reidenouer, & Pantel, 2012).

In psychiatric illnesses, Kuipers (2010) found caregiving roles were associated with an increased level of stress and exhaustion, causing the caregiver’s health to deteriorate. Caregivers were at risk for sleeplessness, post-traumatic stress disorder (PTSD), and depression. The stigma of mental health diagnoses also contributes to additional stress. The author found that many families go through the grieving process with a psychiatric diagnosis, but are inadequately treated (Kuipers, 2010). Additional studies confirm that caregivers are at risk for lower quality of life, social isolation, and higher perceived burden in the psychiatric caregiving population (Hayes, Hawthorne, Farhall, O’Hanlon, & Harvey, 2015; Stanley, Balakrishnan, & Ilangovan, 2017). A study of a non-disease-specific group of caregivers found an increased incidence in depression and anxiety, as well as impaired quality of sleep (Phillips et al., 2009). A similar study reports stress, anxiety, and depression as risk factors for caregivers (Khan, Pallant, & Brand, 2007).

Respiratory considerations are incredibly important to PH patients, as caregivers routinely manage dyspnea in the home setting. A population experiencing similar issues would be caregivers of those who are mechanically ventilated. With patients on prolonged mechanical ventilation, employment was a major stressor for caregivers, as well as depression, and general changes in lifestyle. Caregivers of patients in the post-intensive care unit phase are at greater risk for depression and post-traumatic stress disorder (PTSD) (Van Pelt et al., 2007). In patients with tracheostomies, caregivers of those requiring mechanical ventilation felt strain in the one-year follow-up from the original study, indicating that caregiver burden is longitudinal in this instance. They felt the need for additional information about the disease process if the patient
was on nocturnal ventilation therapy, and both the caregiver and care receiver felt they had restrained lives resulting from the illness (Ferrario, Zotti, Zaccaria, & Donner, 2001).

**Summary of the Background**

The above literature points to caregiving as an important aspect of living with acute or chronic disease, and suggests a correlation of outcomes for the patient and the caregivers’ reactions. In addition, it underscores the importance of social support, compassionate and timely communication, psychological needs of the caregiver, inability to care for acute dyspnea in the home setting, and significant risk factors for the caregiver. These risk factors, summarized from above, could give rise to depression, anxiety, PTSD, drug or alcohol use, stress, exhaustion, and sleep deprivation in the caregiver. There are positive aspects to caregiving, including companionship, a sense of reward, and enjoyment. Caregiver reactions have not yet been described in the PH population, and this study aimed to fill this gap in the literature.

**Problem Statement, Significance, Research Question, and Premises**

The purpose of this study was to describe caregiver reactions within the context of PH. This is significant because of the increasing incidence of PH, the severity of the disease, and the paucity of literature on caregivers of those with PH. The overall cost of PH is staggering. Certain PH medications are estimated to range from $36,000 to $93,000 annually (Widemar, 2005). There is also an increase in frequency of hospitalizations, which has recently tripled, with death rates rapidly increasing (Hyduk et al., 2005; George et al., 2014). Cost of hospitalizations and re-hospitalizations is significantly costly for PH patients, making investigation into caregivers’ experiences paramount as a means of avoiding hospitalization. In one study dealing
with WHO group I PH, the cost of initial hospitalization was $30,286, and the average length of stay was 11 days. The average cumulative cost for re-hospitalization was $71,622, with an average length of stay of 24.5 days. The mean one year total PH-related hospitalization costs per patient was $101,908. The average total cost was $134,830 for commercially insured patients and $53,039 for patients with Medicare Advantage (Canavan, 2013). Cost of hospitalization is higher among those with PH-related complications as opposed to other illness, with PH costing $61,922 versus $42,455 for other illnesses, and a 14.2- versus 10.2-day length of stay in the hospital, respectively (Burke et al., 2015). There are many unmet needs of caregivers, as evidenced by several risk factors and other issues impacting caregivers in the literature review. There is also a significant shift to caregiving taking place in the home, in addition to a rapidly aging population, contributing to a need for information on informal caregivers. The research question is as follows:

*What is the experience of the informal caregiver of a person with pulmonary hypertension?*

A cross-sectional, qualitative phenomenological design was used, with participant interviews generating data for analysis. One premise of this study was that this population encounters some amount of caregiver burden or strain, as those with PH require much care in many facets of living. These include, but are not limited to, finances (due to the cost of PH medications), time spent caregiving and away from employment, sleep deprivation, and psychological and emotional strain, both in dual role strain and emotional stress from witnessing care receivers’ struggles.

Another premise was that there are positive reactions to caregiving, including strengthening caregiver/care receiver relationship, and a sense of self-satisfaction, pride, or gratification from caregiving. These premises were derived from review of the literature, as well
as anecdotal information gathered from PH support group meetings, message boards, and general interactions with PH patients and their caregivers.

**Conceptual Framework**

The conceptual framework selected to frame the methods and support the research question is the Roy Adaptation Model (RAM). The RAM provides the foundation for structuring the methods and analysis of the investigation into caregiver reactions to acute illness, and explains the process and effect of these reactions.

![Figure 1: RAM (Roy et al., 2009)](image)

The *stimuli*, or anything that could provoke a response, contain the *focal stimulus* (the immediate stressor- PH), the *contextual stimuli* (the other circumstances affecting the individual), and *residual stimuli* (the social support or outside factors that could impact the caregiver). The stimuli have a direct effect on the *coping processes*, generally how the person internalizes the stimuli. These *processes* are expressed in the *mode of adaptation*, which consists of adaptive (positive) and ineffective (negative) responses. The *modes* vary according to type of adaptation.
The first, physiological mode, consists of basic physical needs. The self-concept mode involves the beliefs a person holds about himself or herself, in addition to how he or she is perceived by others, which would direct his or her behavior. The role function mode is important to the model’s central theme of the person as an integrated whole. This mode specifically incorporates the psychosocial aspects of the whole person, and is the knowledge of who a person is in relation to others. The incorporation of societal expectations is also integrated into this mode. Lastly, the interdependence mode describes close relationships with others and the ability to give and receive love and respect (Roy, 1984; Roy, Whetsell, & Frederickson, 2009). The overall responses are either adaptive or maladaptive, which ultimately feed back to the stimuli phase. If the informal caregiver is responding in a maladaptive manner, this reaction contributes to the environmental stimuli, which ultimately impacts each of the modes of adaptation.

In application of the RAM to caregiver reactions in PH, the care receiver is the focal stimulus. Contextual stimuli include but are not limited to finances, caregiver health, time constraints, employment, housing situation, and geographic location. Residual stimuli represent support among family or friends, as well as support groups. The environmental stimuli variables were measured with the use of demographic forms, and the qualitative interviews. The coping processes are generally the same as described by Roy-physiologic (regulator) and cognitive processing of the caregiver experience. Within the PH dyad, the caregiver internalizes stress as either stability- maintenance of the norm- or change (stabilizer versus innovator, respectively) (Roy, 1984).

The modes of adaptation provide for the most important application of the RAM to the caregiver experience in PH. The physiologic mode incorporates the health of the caregiver, as reported by the caregiver. Newman (1997) suggests incorporating fatigue and lack of sleep due
to caregiving demands while applying caregiver reactions to the RAM, which will be collected in the demographic form (Newman, 1997). The *self-concept mode* includes the emotional health of the caregiver, threats to the caregiver’s self-concept, anxiety, depression, and guilt—information that will be captured in the qualitative interview and in the demographics form. *Role function* also incorporates some feelings of guilt, or perceived “duty to care.” Dual roles, such as significant other and caregiver, are often difficult for this population, negatively affecting adaptation. *Interdependence* is social support and relationship quality for the caregiver, which was examined within the interviews. The inclusion of the above modes of adaptation will ultimately dictate if the caregiver reaction is adaptive or maladaptive, which could potentially feedback to the original stimuli stage of the RAM. As this proposed study is not interventional, the purpose is to describe the caregivers’ reactions and not to test interventions to modify adaptive or maladaptive reactions.

**Conceptual Definitions**

For the purposes of this study, “pulmonary hypertension” was defined as a pulmonary artery pressure of greater than 25 mm Hg at rest as diagnosed by echocardiogram or right heart catheterization. Caregivers who are unsure of specific diagnostic testing, but endorse PH symptoms and a prior diagnosis from a clinician (defined as dyspnea on exertion, shortness of breath, syncope, and edema) were included in the sample (Steinbis, 2008). The patient with PH was managed primarily in the home by the caregiver.

“Caregiver” is defined as a person with no medical background who is primarily responsible for some aspect of care in regards to a PH care receiver. “Caregiver reactions” is defined as “time devoted to caregiving, difficulty of caregiving tasks, and overall perceived caregiving distress”, as well as positive caregiver reactions (e.g., high esteem and pride in
Inclusion of positive aspects of caregiving will also be noted in participant interviews.

**PH Operational Definition and Semantics**

Caregivers were included in this study if their care receiver had a diagnosis of PH from a right heart catheterization or echocardiogram from a cardiologist or pulmonologist. Caregivers of those with highly suspected but unconfirmed PH, or a previous diagnosis of PH, also were included in the sample. Severity of PH will be measured with the NYHA functional classes, the current accepted clinical standard for functional status in PH. Etiology of PH was defined by the WHO groups I-V (Hoeper et al., 2016; Simonneau et al., 2004), and collected within the demographic characteristics of the PH caregiver for analysis (Appendix II). It is important to note that not only is there a large time period between exhibition of PH symptoms and diagnosis (Brown et al., 2011; Traiger, 2007), but also some confusion related to terminology. Many times, “pulmonary hypertension” (PH) is used interchangeably with “pulmonary arterial hypertension” (PAH). “PAH” is usually exclusively used in the literature for WHO group I patients, or those with idiopathic pulmonary hypertension. For the purposes of this proposed study, “PH” was used as more of an umbrella term to incorporate all WHO group etiologies of pulmonary hypertension. The rationale for using a broader term was to make the caregiver’s experience in filling out the demographic form slightly easier, as they should not be expected to know the etiology of the PH.

**Sampling and Research Design**
The target population was community-dwelling, lay adult caregivers of those with PH. Inclusion criteria were: over the age of 18, able to speak, understand, and read English, and took primary care of a person known to have a diagnosis of PH in the home setting. Criteria for diagnosis was not stringent, as the settings in which caregivers were recruited was primarily accessed by those with confirmed PH. The caregiver had to be certain beyond reasonable doubt that their care receiver had a diagnosis of PH from a specialist. If the caregiver was able to recall specific right heart catheterization results, this information was collected, but not required. Caregivers of those within all WHO group etiologies and NYHA functional classes were included. Individuals excluded were newborns with persistent pulmonary hypertension, non-English speakers, minors, and bereaved caregivers.

The sample was recruited from the Boston PH support group, and the Pulmonary Hypertension Association (PHA) online discussion board for caregivers. Within the support group, caregivers were recruited using flyers with a summary of the study and contact information, and the group leader disseminated information about the study to those who could not attend. The study was introduced at the beginning of the support group session. Informal caregivers were given the option to participate in a semi-structured interview (Appendix C) at a later time via telephone or in a quiet place convenient to the caregiver. The care receiver was not present for the semi-structured interview due to potential bias. The participant was informed of this, risks and benefits, as well as typical length to complete the interview to make arrangements for care, if needed. Participants were informed that interviews would be audio recorded, and they can opt out of audio recording if preferred. No participants opted out of audio recording.

Within the PHA message board (http://www.phassociation.org/DiscussionBoards), a moderator was contacted to add an announcement of the study to the caregiver’s message board.
with contact information to conduct a semi-structured interview via telephone, Google Hangout, or Skype (using a secure connection; depending on participants’ preferences), with audio only. All potential participants were notified that the interviews would be audio recorded for transcription purposes, and again could opt out of this recording. Participants were given as much time as needed to complete the interview, with probes used for further exploration of certain concepts as needed.

The sample consisted of seven informal caregivers, during which time data saturation was reached. One caregiver’s wife had died three days prior to the scheduled interview. This individual was included, with permission from Northeastern University’s Institutional Review Board, despite caregivers with deceased care receivers being exclusion criteria. The individual was included because he was a caregiver so recently. The overall sample had an average of 63.1 years of age, with a range of 54-71, and a standard deviation of 7.17 years. Most of the sample was female and retired, and the majority were highly educated, holding post-baccalaureate degrees. The care receivers were mostly WHO group I, NYHA functional class IV individuals, receiving continuous oxygen. The majority of caregivers also had health problems, themselves, with the most frequent problem being hypertension. See the tables below for more demographic information on the sample.
Table 1

*Care Receiver Characteristics*

<table>
<thead>
<tr>
<th>WHO Group</th>
<th>% of sample</th>
<th>NYHA</th>
<th>% of sample</th>
<th>On Oxygen</th>
<th>% of sample</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>57.1</td>
<td>2</td>
<td>14.3</td>
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</tr>
<tr>
<td>2</td>
<td>14.3</td>
<td>3</td>
<td>28.6</td>
<td>No</td>
<td>42.9</td>
</tr>
<tr>
<td>5</td>
<td>14.3</td>
<td>4</td>
<td>57.1</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Table 2

*Caregiver Characteristics*

<table>
<thead>
<tr>
<th>Gender</th>
<th>% of sample</th>
<th>Education</th>
<th>% of sample</th>
<th>Work</th>
<th>% of sample</th>
<th>Caregiver</th>
<th>% of sample</th>
</tr>
</thead>
<tbody>
<tr>
<td>Female</td>
<td>71.4</td>
<td>High School</td>
<td>14.3</td>
<td>Full time</td>
<td>14.3</td>
<td>No Issues</td>
<td>33.3</td>
</tr>
<tr>
<td>Male</td>
<td>28.6</td>
<td>Some College</td>
<td>14.3</td>
<td>Part time</td>
<td>14.3</td>
<td>Hyperlipidemia</td>
<td>11.1</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Bachelors</td>
<td>14.3</td>
<td>Retired</td>
<td>57.1</td>
<td>Tendonitis</td>
<td>11.1</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Masters</td>
<td>42.9</td>
<td>Disabled</td>
<td>14.3</td>
<td>Hypertension</td>
<td>33.3</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Doctorate</td>
<td>14.3</td>
<td>Disc Issues in</td>
<td>11.1</td>
<td>Back</td>
<td></td>
</tr>
</tbody>
</table>
**Human Subjects Protection and Institutional Review Board Approval**

This study was approved by Institutional Review Board (IRB) at Northeastern University. All communications online were secured using SSL encryption and secure connections, and all data were stored on a password-protected computer within a locked office. Data collected were stored after completion of the study for possible corrections in a secure file for up to one year after study completion. No identifying information was collected, including IP addresses. Participants were able to start or stop interviews at any time, and were provided written informed consent prior to participation. Additionally, interviews were audio recorded and transcribed with explicit permission from participants. These audio recordings were promptly transcribed by hand and then deleted, with transcriptions being stored on a password-protected computer for one year after completion of the study.

**Data Collection and Analysis**

Semi-structured interview data were collected until data saturation was reached, with a sample size of seven. At the beginning of the interviews, Moustakas recommends engaging in an Epoche process, in which judgments about the existing reality cease to exist—a means of removing interviewer (and interviewee) bias, an important exercise in reflexivity. Before interviewing, I have logged any potential bias (e.g., care must be burdensome) as a means of recognizing and attempting to remove bias. Moustakas clearly believed that it is impossible to completely separate oneself (and thus, potential bias) from the data, as the researcher is the instrument, but it is extremely important to be able to identify bias (Moustakas, 1994). Care receivers were not present as a mean of alleviating bias within the interviewee population. A semi-structured interview guide (Appendix C) was used, although at times appropriate probes for
more information regarding an important topic were utilized. Data were then coded utilizing NVivo 11.4 software to form codes in a phenomenological manner using Moustakas’ modification of the Stevick-Colaizzi-Keen method of reading for what was experienced, how it was experienced, how the data become a whole, and finally how it concludes. Moustakas utilized this methodology while exploring loneliness in the decision of cardiac surgery to repair his daughter’s heart defect. He used his own experience to explore and comprehend others’ experiences, which eventually culminated into a more holistic method of collecting and interpreting data. His is a heuristic method; that is, an approach to problem solving or method of thinking that will deepen understanding of phenomenon or create a practical solution. Additionally, he calls for the researcher to become an instrument for data collection, suggesting that transcribing is pivotal to understanding the phenomena in question. The investigator personally transcribing each interview is paramount. Moustakas also addresses bracketing, or reducing bias in qualitative research. He argues that investigators are unable to separate themselves from the human experience, thus making it tacit, intuitive knowledge. While it is important to not make judgments based on this subjective human experience, it is important to recognize life experience is impossible to bracket prior to starting qualitative research, an idea important to heuristic inquiry (Kenny, 2012).

The principal investigator (PI) personally transcribed each interview verbatim. The transcription was then coded by the PI and two graduate assistants in two cycles, specifically for vivid experiential descriptors that Moustakas refers to as the “meaning units” of the experience. Coding took place with the intent of developing patterns, because repetitive actions and consistency of information lends itself to more rigorous findings with greater relevance to the actual human experience being described (Miles, Huberman, & Saldana, 2013, pp. 3-6). A
A codebook was generated and contained a master list of the codes with a brief description of each, as maintaining this list could potentially lead to further reorganization of codes and/or themes.

Coding was completed in two cycles. First cycle coding consisted of In Vivo and Process coding, in which participants’ own words was used to code information (In Vivo coding), and comprehensive gerunds (Process coding) were used to describe a phenomenon within the interviews. These methods provide more action-based descriptors that are appropriate and illustrative of a concept such as caregiving with PH. Process coding, while appropriate for nearly all qualitative studies, is particularly useful in “routines or rituals” of human life, which is clearly applicable to assisting in daily care. Searching for a consequence of an action is also part of process coding, fitting well with a goal of describing caregivers’ reactions. In vivo coding, otherwise known as “literal coding” or utilizing verbatim phrases, was also used to emphasize the importance of the participants’ voices and literal phrasing. Codes also overlapped in first cycle coding, necessitating use of Simultaneous coding, where two or more codes are used for one datum. Transition to second cycle coding was assisted with a word frequency cloud to examine a draft-form of the frequency of language/code selection, and codes were examined to see if they could be split or lumped under another code with similar meaning (Miles, Huberman, & Saldana, 2013, pp. 105, 111.).

As part of second cycle coding, these preliminary codes were organized in such a way that they cluster into patterns based on the categorical or thematic incidence of codes. Hatch (2002) describes this as organizing and reorganizing codes according to similarities, differences, frequencies, sequences, correspondence (i.e. happening in relation to other events), and causation (i.e. one incident apparently causing another) (Hatch, 2002; Miles et al., 2013). Then,
overarching themes were derived from the reorganization of codes (Creswell, 2006; Moustakas, 1994; Miles, Huberman, & Saldana, 2013, pp. 15).

These themes were obtained using second-cycle Pattern coding, or creating categorical groups of similarly coded data, a method especially useful with large amounts of data. Each theme had a verbatim example from the interviewing process that demonstrates the “texture” of the experience (Moustakas, 1994), a method Moustakas called creating “exemplary portraits” of each theme (Kenny, 2012, pp. 9). No more than ten quotes were selected per theme, which were ordered according to relevancy (Miles, Huberman, & Saldana, 2013, pp. 274). This entire process was described by Pinchbeck (2007) as pattern perception in which the researcher is drawn to “meaning units” that relate to the research question. These units adequately describe the phenomena and can “speak” to each other and have meaningful synthesis when combined (Pinchbeck, 2007). These data were managed using NVivo in addition to Quirkos for data visualization.

Credibility and dependability of data were upheld in taking from Moustakas’ modification of the van Kaam analysis. Member checking, or confirmation of themes, was sought from PH caregivers to ensure credibility (Bloomberg & Volpe, 2015). It is important to confirm the research findings with the participants, and to give them a chance to clarify any themes or make a note of information erroneously omitted. A confirmatory message board post with preliminary results was posted for the participants recruited online, and a general announcement was utilized for those recruited via the support groups, posted in a high-traffic area in the support group meeting place. A request for feedback was included along with these preliminary results to assure validity of findings, however, specific participant contact information was not stored to increase confidentiality. Individual participants were not contacted,
and a general non-specific posting was utilized. One individual responded and confirmed all themes were accurate, except for the first overarching theme, as her daughter had multiple other illnesses prior to the development of PH. Dependability was ensured with frequent journaling throughout data analysis, and the maintenance of a codebook.

**Findings**

After second cycle coding, several themes emerged. In addition to the overarching themes, sub-themes emerged from the verbatim transcriptions of the interviews. It is important to note that these themes were presented chronologically to become somewhat of a narrative. They are as follows:

**Initial Diagnosis is Overwhelming**

In the first overarching theme, the initial period of diagnosis was particularly burdensome for caregivers, described frequently by multiple participants as life-altering. In the majority of interviews, the participants discussed this time period in their lives without prompting almost immediately. Subthemes include overwhelming initial diagnosis and a long period of time between initial symptoms and diagnosis. Many caregivers sought out information on the internet (not the PHA message board) and they found this unhelpful due to reporting of grim prognosis timing. As far as diagnosis, one participant sums up this concept by stating, “In terms of the worst diagnosis you can get… It’s more than you can take.” Many participants wait a long time for diagnosis. Only one caregiver did not feel this was the case because her son was so young and previously healthy.
Diagnosis was difficult but conclusive. One participant described an expanse of time from onset of symptoms to diagnosis as being due to using comorbid conditions to explain early PH. “I really noticed a difference when we were on a cruise in 2014. And before that she was complaining about shortness of breath. She was also obese, so, you know, they would always say, ‘well, you’re overweight, that’s why you’re short of breath, why you can’t walk that far’.” This participant described initial symptoms as initially attributed to comorbid conditions, thus prolonging the time to diagnosis. A PH specialist was necessary for all of the participants to obtain a diagnosis.

Becoming Clinically Literate

The second overarching theme, immediately following the diagnosis, is becoming clinically literate immediately. One participant described it succinctly as “rapidly becoming a medical family.” Subthemes include avoiding exacerbation, each case of PH is different, dealing with side effects from medications, preparing with either helpful or unhelpful medical professionals, and the need for increased awareness of PH. Many caregivers quickly learned that fluid overload rapidly leads to an acute exacerbation, making them specifically point out this sign as a need for increased diuresis. Many also made a point of mentioning each case of PH is different, so it is important to have individualized education for caregivers. Side effects of medication were frequently referenced by two participants in particular. Many of the care receivers were on treprostinil, which is administered subcutaneously, and many had such serious site irritation that it impeded everyday life for the person with PH and severely impacted caregiving. One of these participants experiencing side effects described it as far worse than when she went through radiation for breast cancer.
As far as preparation and support by medical professionals, many caregivers felt as though their rapport with their PH specialist enabled them to call with any questions and be confident that their questions would be answered in a timely manner. Additionally, the nurse available for treprostinil administration care was an excellent resource. The participants stated it was imperative to have a PH specialist, as a general practitioner would not be very well-versed with such a niche disease as PH. It was also important to the caregivers to increase knowledge and awareness about PH because many of the medical professionals in their local hospitals were unfamiliar with the disease and many of the people in their everyday lives were also unfamiliar. In the example of the participant with the site irritation due to treprostinil, there was regret that it was so difficult to explain to her friends her severity of illness, as she did not outwardly appear ill beyond the oxygen tank. The site irritation was debilitating and painful, and she frequently had to skip social activities due to the pain and infection risk.

**Embracing A New Normal**

This overarching theme highlights the importance of quality of life over quantity and the fact that PH is isolating. Embracing the “new normal” is the first step in coping for these lay caregivers. This theme is about living life with modifications, coping, and some of the drawbacks that come with that. Advocacy was a major concept within this theme and the new role as advocate that caregivers had to assume. Many caregivers felt unprepared for this role by their prior education and training, with one participant saying, “it’s one thing to lead a project where you’re installing 10,000 computers and there’s a little glitch, and another thing when it’s your sister, and she’s scared and worried that the site is bleeding, and it’s Easter, you know… It’s just, it’s different.”
Life is described as changing drastically for the caregivers. There is uncertainty about the future where there was a once-healthy individual. Presence of pain is also a new normal for one caregiver, as her care receiver experienced deep bone pain. This phenomenon is described by one caregiver very well. “The numbers are good, but her lifestyle… I mean when they say that this kind of thing changes [your life]– it’s a new normal – boy, that is the truth. It’s a degraded normal, but you are alive, so that’s the good news.”

PH is described as isolating, because many people are unfamiliar with the disease. Caregivers described lay people as less understanding because PH knowledge is not common knowledge. One participant describes this phenomenon as, “you end up in kind of an isolated position in life, because friends have problems, you know, and sometimes they’re medical problems, but not a lot of people who I know have a chronic [disease]– other than maybe diabetes, but a chronic severe…rare health problem.” Her care receiver has felt this lack of understanding from others, as well, “at times she’s said she’d like to just have cancer back because it’s more known and people know about it, so if you tell people you’re dealing with that they can sympathize. Pulmonary hypertension? No one knows about that.”

**Caregiving Creates Stress**

With the continued coping comes stress. In this sample, informal caregiving has created a fair amount of stress; “…it was extremely stressful. It was heartbreaking. It took a lot of time. I mean, there was a lot of times when she was sleeping, I would sit there and just watch her sleep.” Between physician’s appointments, which were quite frequent for this group of caregivers, the uncertainty about the future, dealing with pain and side effects from medications, caregiving and the general management of care had taken a physical and emotional toll. Also important to note
within this overarching theme, there were other family members and many other older adults who needed care. One individual is a dual caregiver to her sister with PH and her ill mother. Another caregiver in this study had three other children who required constant care. There was a general “worrisome” feeling described, with caregiving being emotionally difficult and at times a struggle. One man, who had lost his wife three days prior to calling and inquiring about the study, encapsulated this “worrisome” concept very well. He stated, “sometimes she would get to sleepin’ so deep that it was... I would have to wake her up, you know, and get her to sit up for a little bit. And she’d be… you know, I’d try to wake her up, and she’d be all, ‘I’m alright, I’m okay!’. I’d say, ‘no you’re not!’. It would take a long time. So I would get her to wake up, to sit up for me for about ten minutes or so, and I’d let her get back to sleep. I would sit there and literally watch her sleep for hours. Just... watching...” Management of medications, side effects of medications, assessing for an acute exacerbation, time constraints, sleeplessness, and emotional turmoil were conditions that created stress for these participants.

**Getting Support and Self-Care**

In order to mitigate the stress and isolation, informal caregivers suggest seeking out support and finding a way of improving their own quality of life with self-care. Support is important for both the caregiver and care receiver. One concept that consistently came up with all participants was general advice: “do not go through it alone.” It was an interesting juxtaposition, however, because many caregiver/care receiver dyads were not able to attend in-person PH support group meetings and the Boston PH support group meeting appear sparsely populated. Many participants explained that this was because logistics are particularly difficult. Most meetings are held at major metropolitan hospitals and parking became an issue. In addition
to this, traveling with oxygen was particularly difficult, especially in the city where one may not have an oxygen condenser, and if one must travel from far away. Travelling without an oxygen condenser makes oxygen tanks heavy and burdensome, and estimating how much oxygen will be used is difficult in planning travel.

As a consequence of travelling being so arduous, the PHA message board was utilized more as a means of support due to convenience. Almost every participant referenced the PHA website as both a resource and means of support from peers. The resource was particularly helpful to one individual who had to do all of the researching on her own time as her care receiver found the information online too disturbing. Finding support with friends and loved ones was important, as well. “You can’t go through it alone. You have to depend on your friends or a spouse. Try not to go through it alone.” Support is essential to continued coping. Self-care is also very important. Some participants spent time with their friends, some meditated, some practiced yoga, some engaged in retail therapy, and one participant found stress-relief through a regular pedicure. Regular exercise also increased the caregivers’ abilities to cope.

**Improving Relationship Quality**

One overwhelming positive aspect of caregiving with every participant was the overall improvement in relationship quality between the caregiver and the care receiver. Many of the participants admired their care receivers for being so resilient. “My wife, I’ll tell you what, I mean, I was… the doctors were just amazed. There’s things that happened that only a few of us know or have experienced. She wouldn’t even tell me about it at first. You know, for a long time. She thought she was crazy or something. I said, ‘no… just tell me. Just tell me about it.’ I said,
‘Wow! It’s just pretty neat’.” Many were caregiving for love, and advised to do activities with each other, not to show frustration, and provide emotional support.

Caregivers were “honored and happy” to provide care. Reinforcing caregiving for love, honor and happiness, one participant says, “…but at the same time, you know, I loved her with all my heart. She had a heart of gold (cries). She would do anything. My advice is to be patient. Caring. Loving. Supporting. Do everything they can for them. You know?”

In addition to providing caregivers and care receivers common ground for discussion, the seriousness of the disease also brought out more frank discussion between the caregiving dyads. One caregiver noted that she would read particular sections on the PHA website, and be able to discuss with her sister the next day. Multiple participants said that their relationship was deepened by the PH, as they have learned more things about their partners and have gotten more direct, honest answers due to the seriousness of PH. They have learned different ways to be supportive of their care receiver and have been “lucky” to have spent as much time as they have with their care receiver.

**Summary of the Themes**

The above findings fit well with the Flattery et al. (2005) findings in the lived experience of PH. Their study did not contain any caregivers, but found two overarching themes: holding back (fear, anticipating that symptoms will worsen, side effects of treatment), and redefining life (uncertainty, restrictions on daily activities, coping). “Holding back” and its subthemes appears consistent with the initial diagnosis being overwhelming, and caregiving creates stress. “Redefining life” is consistent with becoming clinically literate, embracing a new normal, and utilizing support and self-care. This also seems to agree with McDonough, Matura, and Carroll’s
(2011) qualitative study, in which investigation of the PH patient experience found overarching themes of holding back (fear, anticipating that symptoms will worsen, and side effects of treatment), and redefining life (uncertainty, restrictions on daily activities, and coping). However, improving relationship quality is a novel finding unique to this study on caregivers.

Additionally, these findings fit well with the Roy Adaptation Model (RAM), which guided the methodology and semi-structured interview content. The environmental stimuli continue to be PH and other circumstances affecting the individual caregiver. The most pertinent points of the model are in the Modes of Adaptation in the RAM. The physiological mode displays itself in all of the themes, such as the stress created by caregiving, the initial diagnosis being overwhelming, and the level of preparedness giving medications in the home. The self-concept mode is evident by feelings of support by healthcare professionals and usage of stress-relief activities. Role-function and interdependence is displayed by the successful caregivers sharing advice, “do not go through it alone,” with fellow caregivers. These two modes are also explicit in the overall improvement of relationship quality between the caregiver and care receiver. Ultimately, management of the above modes feed back into the environmental stimuli, and overall caregivers are successful, adaptive, and resilient, as evidenced by this group still providing excellent care (Roy, 1984).

Figure two represents a word cloud of word frequencies. Every one of the participants was extremely pleased to see that research was being done on the PH caregiver, with one caregiver saying, “I would be the first to say that [caregiving] is the most under-acknowledged aspect of this whole thing.”
Although data derived from this research are very important and salient to those with PH and their caregivers, there are limitations. The study is the first step to future quantitative studies that may provide evidence of need for funding or assistance for the caregiver population. Further, the sample of participants recruited, although from multiple settings, is limited to those in the Boston area or globally to those owning and able to use a computer and the internet. This makes
the data non-generalizable to other populations, and can potentially exclude those without access to a computer. Selection of a convenience sample also limits generalizability of this study.

Although the use of the PHA message board partially ameliorated geographic limitations, many of those with PH were just too ill for their caregivers to spare time for these interviews. Anecdotally, while working on other inquiries into PH health-related quality of life, many of those with PH were too ill for traveling, and it can be inferred that their caregivers would not be able to spend time participating in an interview. Many times, travel to the support group is very taxing for both PH patient and caregiver, potentially making the caregiver less likely to participate in this study. Thus, this study may have inadvertently excluded those with very severe PH, or NYHA functional class IV, on continuous epoprostenol. Ideally, availability of responses from the PHA message board broadens the sampling approach to include those caregivers whose care receiver is too ill to participate in person.

The sample was largely homogenous—mostly females in their 50s-60s with an education level greater than high school. Most of the sample was retired, so the time constraints a full-time worker may experience are not fully characterized in this study. At the same time, the inclusion of retirees may have amplified the financial burden for those on a fixed income. The findings are not necessarily transferrable to the male caregiver, which was one phenomenon that would be interesting to capture. The two males’ interviews included in the study were not significantly different from the females’ except for being shorter and slightly less descriptive. Those with lower levels of education also may experience caregiving differently, as well as those significantly younger or older than the average age in the sample.

Roy described adaptation or maladaptation as a response to the modes of adaptation modulated by the coping processes of the caregiver. Although it would likely reveal very
interesting information, those who have responded in a maladaptive manner may not be included within the targeted sample. Those with a maladaptive reaction most likely would not be actively caregiving, which is an exclusion criterion within this study. Exploration of those unable to tolerate caregiving for those with such an acute-on-chronic disease state would be a very interesting topic for further study, but is outside of the scope of this particular study.

**Conclusions, Recommendations, and Implications**

The paucity of literature about adults with PH and their caregivers supports the need for continued study on informal caregiving. With the current climate in healthcare reform and potential to transition to the American Health Care Act of 2017 (H.R. 1628), the Congressional Budget Office recommendations indicate that the number of uninsured people could increase to 24 million by 2026. The rate of uninsured is estimated to be 18.6%, whereas this rate was projected to be 10% by 2026 under the Patient Protection and Affordable Care Act of 2010. Under this reform, older, sicker, and poorer Americans will likely be paying higher premiums and out-of-pockets costs (Congressional Budget Office, 2017). This could potentially put increased burden on both the caregiver and the care receiver, making research on how to best support the dyad imperative. Potential risk factors for caregivers, severity of disease for the care receivers, and cost of care, as well as potential mismanagement of dyspnea in the home clearly indicate the significance of the problem. Qualitative methods are best suited for this inquiry into the experience of the PH caregiver because of the aforementioned lack of literature on this specific topic as well as a demonstrated need for basic description of the problem to determine the direction and aims of future research in this area.
The findings included six overarching themes, which also had sub-themes. They were as follows: Initial Diagnosis Overwhelming (diagnosis overwhelming, long time from initial symptoms to diagnosis), Becoming Clinically Literate (avoiding exacerbation, each case is different, side effects from medications, preparation by medical professionals, increasing awareness of PH), A New Normal (new normal, PH is isolating, quality not quantity), Caregiving Creates Stress (caregiving is stressful, caregivers are under acknowledged, others need care), Support and Self-Care are Imperative (be supportive as the caregiver, don’t go through it alone, support groups, self-care as stress relief), and Relationship Quality is Improved.

Implications of this study include a need for increased caregiver support provided through the use of the internet. Many of the participants were unable to attend PH support group meetings due to many logistic issues, including travelling with oxygen, parking, and traffic. The PHA website has been an enormous support to these individuals, including a source of stress relief with the initial diagnosis packet answering many questions. Those with PH and their caregivers can receive a brief and easy-to-understand packet of general information, entitled “Envelope of Hope” about PH, as well as a glossary of terms. Typically, the caregivers were initially shocked at diagnosis, but more so by the frequently reported two-year prognosis at diagnosis of PH. The PHA’s informational packet frequently was cited by caregivers as an important initial resource. Although the PHA is supportive, individual participants would have like to see increased programming for the caregiver. One caregiver referenced a PHA-sponsored lay caregiving conference that was helpful, and need for future programming, as there has not been another conference. Another caregiver specifically wanted a connection with another caregiver in a similar circumstance to have for commiseration. Face-to-face regional meetings
that did not take place in major cities were frequently desired for the care receivers because of the difficulty of travelling into the city with oxygen.

Additionally, there needs to be increased support for the informal caregiver by the PH specialist. Many participants were very fortunate to have a good rapport with their specialist, but some individuals would have appreciated a warmer approach that took into account the caregivers’ needs. One individual in particular would have liked to have a “caregiver contact”- a wife of a patient that she could speak to regarding issues that arise and common frustrations of at-home caregiving. There is evidence to suggest that peer support positively affects psychological and physical health outcomes (Dennis, 2003), and social support, coping, and quality of life (Womba, Nijland, Ket, & Kwakkel, 2016). Individual peer support has demonstrated significantly higher self-care in a heart failure population (Riegel & Carlson, 2004). Logistics with oxygen, medication administration, and travelling are issues this caregiver would like to discuss with others who are familiar with caring for their loved one with PH, as well as seeking out peer support.

Informal caregiving is very stressful. Many seek out counseling and some participants were unsuccessful in finding a therapeutic environment to speak with about their stress in caregiving. It would be an important consideration to streamline counseling for both the informal caregiver and the care receiver as a standard of care in PH. The study sample was well-educated, and were able to actively identify the need for professional counseling, as well as navigate mental health insurance coverage. Despite this social capital, they still face challenges. Easier acquisition of such services is essential. PH has been shown to be very isolating and having an outlet to speak with could potentially improve the lives of those with PH and their caregivers.
A major theme found was not going through the process of caregiving alone. Most caregivers felt well supported and well prepared by healthcare professionals, and the availability of the Accredo® nurse for those individuals on Remodulin® was cited multiple times as a very useful service. This model could possibly be replicated by PH specialists for those on oral therapy. Availability of resources could also possibly mitigate some of the stress from caregiving. Also, the time period at diagnosis seems particularly stressful for this population. Coping with this new lifestyle change impacts both caregiver and care receiver, so increased amount of services at time of diagnosis would be helpful. Services may include counseling, acquisition of social services, information about support groups, assistance in attending support groups, information sessions on medications, or even visiting nurse support to possibly mitigate the stress at diagnosis. The sample reported that embracing a “new normal,” meant continuing to live life and resume usual activities. PH care receivers also should be encouraged to resume normal activities in a modified fashion if needed. In all, utilizing these approaches with informal caregivers would utilize early intervention with provision of counseling and peer support, as well as strength based practice, or emphasizing self-determination and assets.

Although it was difficult to recruit for this particular study, a well-powered quantitative or mixed-methods inquiry into the issues concerning caregivers would be very useful, specifically including availability of mental health services, means of coping and support, and self-care methods. Further research on the resiliency of these caregivers would also be helpful to eventually construct an interventional study to improve the lives of both PH caregivers and care receivers. Of note, there were no maladaptive caregivers involved in the study, as maladaptive caregivers would not be providing care. It would be beneficial to study these individuals, if they could be identified, to examine what makes caregiving overwhelming. This information could
also benefit future caregiver interventional studies. Knowledge about the PH caregiver’s experience could lead to future studies that can facilitate more positive caregiving experiences, and likely correlate with better outcomes for both PH patient and caregiver.
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Appendix A- Table 3- WHO Classifications of PH

Table 3

**WHO Classifications of PH**

<table>
<thead>
<tr>
<th>Group</th>
<th>Classification</th>
</tr>
</thead>
<tbody>
<tr>
<td>I- Pulmonary Arterial Hypertension</td>
<td>Idiopathic&lt;br&gt;Familial&lt;br&gt;Associated with:&lt;br&gt;- Connective tissue disease&lt;br&gt;- Congenital systemic-to-pulmonary shunts&lt;br&gt;- Portal hypertension&lt;br&gt;- HIV infection&lt;br&gt;- Drugs and toxins&lt;br&gt;- Other (thyroid disorders, glycogen storage disease, Gaucher's disease, hereditary hemorrhagic telangiectasia, hemoglobinopathies, myeloproliferative disorders, splenectomy) Associated with significant venous or capillary involvement &lt;br&gt;Pulmonary veno-occlusive disease (PVOD)&lt;br&gt;Pulmonary capillary hemangiomatosis (PCH)&lt;br&gt;Persistent pulmonary hypertension of the newborn (PAHN)&lt;br&gt;31.88 (10.29)</td>
</tr>
<tr>
<td>II- PH Related to Left Heart Disease</td>
<td>Atrial or ventricular disease&lt;br&gt;Valvular disease</td>
</tr>
<tr>
<td>III- PH Related to Respiratory Disease</td>
<td>Chronic obstructive pulmonary disease (COPD)&lt;br&gt;Interstitial lung disease (ILD)&lt;br&gt;Sleep-disordered breathing, alveolar hypoventilation&lt;br&gt;Chronic exposure to high altitude</td>
</tr>
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</table>
### Developmental lung abnormalities

#### IV- PH due to Chronic Thrombotic Disease
- Pulmonary embolism in the proximal or distal pulmonary arteries
- Embolization of other matter, such as tumor cells or parasites

#### V- Miscellaneous Group
- E.g. sarcoidosis, histiocytosis X and lymphangiomatosis

Appendix B- Demographics Form

Age:

Gender:

Education Status:
- 8th grade or under
- Some high school
- High school diploma
- Some college
- Associates
- Bachelors
- Masters
- Doctorate

Employment:
- Part Time
- Full Time
- Temporary
- Per Diem
- Retired
- Disabled
- Unemployed

Do you have any health problems?

How much sleep, on average, do you get per night?

Number of hours devoted to care per week:

PH care receiver’s medications:

Are they on oxygen?

PH care receiver WHO group and NYHA functional class (if known):
Appendix C- Semi-Structured Interview

Focal Stimuli/physiological mode

1) Tell me about [care receiver]. How was s/he diagnosed? Please tell me about the diagnosis.

2) How does caregiving for someone with pulmonary hypertension affect your life?

3) How do you feel about giving medications at home?

Self-Concept

4) What about education- do you feel adequately prepared by health care professionals? Please describe the preparation you received from health care professionals. What type(s) of continuing support do you receive from the health care professionals? Do you currently feel supported by health care professionals?

5) Do you utilize any stress-relief activities? Please describe any stress-relief activities you use.

6) Would you consider yourself resilient? Describe your methods of continued coping.

Role-function/ Interdependence

7) Are you involved in any support groups? What is that experience like?

8) What or who do you rely on for resources if you have questions about anything?

9) What advice you would give someone new to in-home caregiving of his or her loved one?

10) Please describe your relationship to the person with PH.
Chapter 5: Summary and Conclusions

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Exploration into caregiver reactions has contributed to the field of informal caregiver research by investigating the impact of a disease state, pulmonary hypertension (PH), on the lay, informal caregiver. The review of the literature suggests there is a certain amount of stress from caregiving and there may be a possible correlation between patient outcomes and caregiver reactions. Additionally, the importance of social support, risk factors for the caregiver, and inability to care for acute dyspnea in the home setting were highlighted. Caregiver reactions are important due to the increasing incidence of PH, severity of disease, cost of care, limited amount of research available, and presence of unmet needs. This is a unique population because these care receivers tend to be female and PH typically exhibits itself as an acute-on-chronic disease state that requires extremely complex care within the home (Matura, McDonough, & Carroll, 2014). The literature also supports existence of positive reactions in caregiving, including caregiving for love and caregiving producing positive esteem (Given et al., 1992), and emotional rewards, personal growth, competence and mastery, reciprocity, relationship gains, and role satisfaction (Lloyd, Patterson, & Muers, 2016). The positive domain is imperative to study as its inclusion gives a more comprehensive, holistic view of informal caregiving.

The first article contains a modified Walker and Avant (2005) concept analysis on caregiver burden, and why it is important to use a more neutrally-connotative word. Burden research has evolved from the 1960s as a largely two-dimensional concept, to gerontology research involving activities of daily living and social support in the 1970s-1980s, to a more multidimensional concept in the 1990s. Positive caregiver reactions were initially studied in 1992 (Given et al., 1992). Caregiver burden continues to be a popular topic as the baby boomer generation began to reach age 65 in 2011, resulting in middle aged adults responsible for caregiving their older adult parents (Centers for Disease Control and Prevention, 2013).
The following chapter explores quantitative instruments to measure the caregiver experience, fulfilling the empirical referents portion of the Walker and Avant concept analysis. The quantitative instruments reviewed included the Bakas Caregiving Outcomes Scale (BCOS), the Caregiver Reaction Assessment (CRA), the Modified Caregiver Strain Index (MCSI), the Zarit Burden Interview (ZBI), the Perceived Caregiver Burden Scale (PCBS), the Multidimensional Caregiver Burden Inventory (MCBI), and the Screen for Caregiver Burden (SCB) (Bakas, Champion, Perkins, Farran, & Williams, 2006; Given et al., 1992; Novak & Guest, 1989; Stommel, Given, & Given, 1990; Thornton & Travis, 2003; Vitaliano, Russo, Young, Becker, & Maiuro, 1991; Zarit, Reever, & Bach-Peterson, 1980). Reliability and validity of each instrument was reported, as well as a short history of the instrument and studies that utilized it. Strengths and weaknesses were explored, but it is important to consider that each instrument is different. An instrument can be selected based on its finer nuances for a common goal of obtaining tangible, objective data on the phenomenon in question.

Following the review of instruments, caregiver reactions in PH is explored. In this qualitative study, the sample consisted of seven informal caregivers with a mean age of 63 years, recruited from Boston-area PH support groups and the Pulmonary Hypertension Association (PHA) message board. Most had a post-baccalaureate degree, and most were retired. The caregivers also tended to have health problems, themselves. Although the sample of participants recruited in this study was predominantly homogenous and female, very useful information was gathered. Many of the care receivers in this sample were quite ill, with the majority receiving continuous oxygen and having a New York Heart Association (NYHA) functional class of III-IV.
Semi-structured interviews were utilized and lasted from 15-60 minutes. Potential bias was logged prior to data collection and analysis and a journal was kept throughout data analysis to contribute to dependability. For analysis, Moustakas’ methods were modified and coded using In Vivo and Process coding. These codes were then second-cycle Pattern coded into patterns to arrive at themes (Miles, Huberman, & Saldana, 2013; Moustakas, 1994; Saldana, 2015) utilizing NVivo 11.4 qualitative data manager, as well as word clouds for frequency of themes.

Findings included six major, overarching themes, which also had sub-themes. They were as follows:

**Initial Diagnosis Overwhelming**

Initially, there is shock at diagnosis and most information available online states a poor prognosis. There was also a long period of time between initial symptom exhibition and diagnosis. In many instances, PH was initially misdiagnosed as a pre-existing comorbidity of the care receiver.

**Becoming Clinically Literate**

Following diagnosis, participants described their families as transitioning, almost immediately, into a “medically savvy” family. The informal caregivers were well versed in symptoms of an exacerbation and in avoiding an exacerbation, and noted that each case of PH is different and could present in different ways. They also described the side effects from medications as being difficult to deal with, and they further described their preparation by medical professionals as being generally satisfactory.
Embracing A New Normal

The overarching theme demonstrates the importance of quality of life over quantity, and the fact that PH is isolating. Embracing the “new normal” is about living life with modifications, coping, and some of the drawbacks that come with that. Assuming the role of advocate and adapting to modifications were some issues facing caregivers. In addition to this, PH is a very isolating disease to caregivers. Many individuals do not know about PH and many with PH may not appear outwardly ill. This makes coping difficult to the point of one participant wishing her care receiver had cancer, or a more well-known disease. PH is also isolating to this group in the sense that it is physically difficult to travel to different places, especially on supplemental oxygen. One participant described a failed trip to a slightly higher elevation that her PH care receiver could not tolerate.

Caregiving Creates Stress

Caregiving did create stress, mostly emotionally, among the participants interviewed in this study. Most participants were very well educated, and retired, so financial stress was not well captured. They did feel under-acknowledged, as PH specialists would tend to focus on the care receiver and not set up support services for the caregiver. In fact, one caregiving participant felt her needs were completely ignored by the PH specialist. Another participant specifically cited the need to talk to another caregiver of a husband with PH, highlighting the importance of peer support as a mean of social support and camaraderie. There are also many others who need care in these caregivers’ lives, and in many instances there was a dual-caregiving situation. One caregiver had both her son with PH to care for, in addition to her other children. Another caregiver had her husband with PH and an older adult parent who was chronically ill. PH can
also be extremely isolating, as many people have not heard of it and do not understand the oxygen requirements, medications, symptoms, and depth of care. One participant said she wished she had cancer again, as other people instantaneously recognize the stress or burden of cancer. PH can also be physically isolating due to difficulties with travelling with oxygen as well as other health issues.

**Getting Support and Self-Care**

Informal caregivers recommended seeking out support systems immediately, even if it is only the PHA message board. Many did not attend support groups due to logistic reasons, but did participate in the PHA message board or with another caregiver in a similar situation. Self-care, be it retail therapy for one participant, exercise, or a manicure/pedicure for another, was imperative to maintaining the caregiving role.

**Improving Relationship Quality**

One extremely positive aspect of informal caregiving was the improvement of relationship quality. Caregivers reported having frank conversations with their care receivers that they otherwise would not have had, and they are able to connect on the common ground of PH-specific care. Caregivers are “honored and happy” to provide care and many caregivers described it as “lucky” that they had so much time with their care receivers. They learned more things about their partners and many described their motivation for caregiving as love.

Limitations included a convenience sample that was mostly homogenous. Most of the sample were females in their 60s with a high level of education. This sample was geographically
CAREGIVER REACTIONS IN PH

Diverse due to recruitment from the national PHA message board, however, transferability of this study is limited to a similar population. Most caregivers had master’s degrees and were retired, so financial burden may not have been adequately captured in this particular sample. In addition to this, maladaptive caregivers were not explored, as they would not be actively caregiving, which an exclusion criterion for this study.

Implications of this study include the need for increased support through the internet, as many individuals were not able to attend in-person support groups due to logistical constraints. Many found the PHA website to be particularly helpful, as well as the diagnosis packet sent by the PHA at diagnosis. Generalized internet searching was not advised by caregivers, as prognosis cited online is dismal. Additionally, more support for the caregiver was needed from the PH specialist, including the need for connection to one-on-one peer support from other caregivers in similar situations. Many caregivers wanted to participate in counseling throughout the caregiving process. Ready and inexpensive access to mental health services would be helpful. There is also a need for future research, to hopefully construct an interventional study to enhance both caregiver’s and care receiver’s embracing of the “new normal” of life adapted to PH.

Assistance for caregivers is imperative, as demonstrated by the National Institute of Nursing Research (NINR) strategic plan (NINR, 2016). The amount of care receivers is growing, with roughly 45% of the population having at least one chronic disease. There is an estimate of 22-26 billion hours of caregiving given annually. Cost of caregiving, with opportunity costs of $522 billion annually, is expensive. Caregiving has many implications for the individual caregiver, as well (Chari, Engberg, Ray, & Mehrotra, 2015). Caregivers have experienced stress, depression, anxiety, a lower quality of life, and financial burden as a result of caregiving.
There are also positive aspects to caregiving, such as esteem or pride in caregiving, and the strengthening of the relationship with the care receiver. The caregiver’s experience is multidimensional, and can be both positive and negative. Further interventional study is required in this population, as demonstrated by the needs of the participants in this qualitative study, the severity of disease, the difficulty of medication management, and the cost of caregiving. Although this sample of PH caregivers was incredibly resilient and able to access resources to enable their provision of care, it is imperative to continue studying this phenomenon for other informal caregivers who may not have access to the same resources. Interventional study could uncover better means of coping for these PH caregivers, leading to more positive caregiving experiences.
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