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Intractable Epilepsy: Does Perception of Epilepsy Stigma Affect Levels of Caregiver Burden?

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INTRACTABLE EPILEPSY: DOES PERCEPTION OF EPILEPSY STIGMA AFFECT LEVELS OF CAREGIVER BURDEN?

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A DISSERTATION

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INTRACTABLE EPILEPSY: DOES PERCEPTION OF EPILEPSY STIGMA AFFECT LEVELS OF CAREGIVER BURDEN?

BARBARA HANSEN

MEDICAL SOCIOLOGY

ABSTRACT

Objective: The purpose of this study is to explore the relationship between affiliate stigma and the levels of burden experienced by individuals caring for family members with intractable epilepsy and to examine how levels of burden may vary between those caring for children and those caring for adults.

Methods: This cross-sectional, quantitative approach utilized a self-administered survey offered to caregivers of family members with confirmed diagnoses of intractable epilepsy in a southern state. Caregiver burden was measured using the 30-item Carer's Assessment of Difficulties Index while levels of perceived stigma were assessed using a six-item scale. Demographic data concerning both the caregivers and their family members were obtained as well. Four nested OLS regression models were estimated using Stata 13.1 software package.

Results: Demographically, respondents (N=136) were primarily female (75%), White (83%), and married (69%) with an average annual household income of just over $80,000. The family members with intractable epilepsy ranged in age from 2-82 years and were evenly split between males and females. Each of the OLS models yielded a positive and statistically significant association ($p<.001$) between caregiver burden and perceived stigma. Additionally, the age of the family member with epilepsy moderated ($p<.05$) the effect.

Conclusion: Results from this study strongly support the argument that as levels of stigma perceived by a caregiver increases, their perception of burden also increases. As well, the relationship between stigma and burden is stronger when the caregiver's family member is an adult. It is important for medical and social service providers to take these results into consideration when determining services to best meet the needs of families.

Keywords: caregiver burden, affiliate stigma, intractable epilepsy, epilepsy stigma

DEDICATION

For my late parents, John F. Hansen and Charlotte Risley Hansen

The fog comes
on little cat feet.
It sits looking
over harbor and city
on silent haunches
and then moves on.

-Carl Sandburg
Fog

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LIST OF ABBREVIATIONS

AED	antiepileptic drug
AFDC	Aid to Families with Dependent Children
ANOVA	analysis of variance
CADI	Carer’s Assessment of Difficulties Index
CAMI	Carer’s Assessment of Managing Index
CASI	Carer’s Assessment of Satisfaction Index
CBD	cannabidiol
CIE	Caregiving in Epilepsy
DV	dependent variable
EEG	electroencephalogram
FDA	Food and Drug Administration
ILAE	International League Against Epilepsy
IRB	Institutional Review Board
IV	independent variable
KMO	Kaiser-Meier-Olkin
OLS	ordinary least squares
PCA	principal components analysis
PWE	person(s) with epilepsy
QOL	quality of life

SDH	social determinants of health
SSD	Social Security Disability
TANF	Temporary Aid to Needy Families
VEEG	video electroencephalogram
VIF	variance inflation factor
WHO	World Health Organization

Chapter 1

INTRODUCTION

The first thing that may spring to mind when someone mentions epilepsy might be an image of a man suddenly falling to the floor convulsing, eyes rolled back, and frothing at the mouth while helpful passersby shout for a wooden stick to shove in his mouth to keep him from swallowing his tongue. Although media representations of epilepsy in movies and television have changed somewhat over the years, they still tend to depict people with epilepsy (PWE) as scary, unpredictable, dangerous, and out-of-control (Kerson et al. 1999). This does little to inform the general public about the varied nature of this disorder and contributes to the perpetuation of the stigma associated with epilepsy. PWE are treated differently and are often avoided or excluded from social activities (Dilorio et al. 2004). In addition to being unpredictable or dangerous, negative stereotypes also include the ideas that PWE are unreliable, should not procreate, are unable to maintain work responsibilities, or unable to function in social roles. They are sometimes avoided by others due to fears of epilepsy being contagious or because of limited knowledge about what to do during a seizure, or the belief that the PWE was likely to die during a seizure (Jacoby et al. 2004).What is not commonly understood is that there are several different types of seizures and some PWE face significant physical and intellectual challenges that interfere with their activities of daily life (Kwon and Brodie 2000). Care needs in epilepsy can be as simple as providing transportation or can include more complex activities such as feeding, bathing, and changing diapers for those with severely debilitating brain injuries. Some PWE do not respond well to medications or surgery and, as years of seizures take their toll, levels of need may increase. With the majority of this care being provided by family members, the level of burden on caregivers can be great (Gibson 2014).

There is evidence of stigma associated with epilepsy (Jacoby and Austin 2007; VanStraten and Ng 2012; Gibson 2014), and other literature supporting the notion that caregiver burden in epilepsy is widespread (Nolan et al. 2006; Van Andel 2009; Karakis, et al. 2014). However, there is little information on how stigma itself might be associated with caregiver burden. As well, different challenges may arise for caregivers of pediatric patients than for adult patients. The relationship between stigma and caregiver burden may vary between these two age groups, yet few studies have proposed what these possible variations might be. This study aims to: 1) examine the relationship between a caregiver's perceived level of stigma and perception of burden and 2) assess what role patient age might play in this relationship.

Motivation for the Study

Epilepsy, a common chronic neurological disorder, is characterized by one or more types of seizures ranging from minor "auras" to grand mal seizures, which are often portrayed on television with convulsions and unconsciousness (Shorvon 1996; Kerson et al. 1999). Treatment for epilepsy is usually effective and typically involves one antiepileptic drug (AED) and then discontinuation of the drug after two years if the patient is free from seizures (Callaghan, Garrett, and Goggin 1988). However, approximately 30% of PWE have intractable or refractory seizures that are not relieved by medications or other treatments ((Kwan and Brodie 2000). Affecting both children and adults, intractable epilepsy adversely affects the physical and mental health of both children and adults and interferes with normal social functioning (Devinsky 1999). In a 2005 analysis of South Carolina data (MMWR 2005), almost half of those studied who reported ever having epilepsy and over 85% of PWE with active seizures in the previous three months (indicative of intractable epilepsy) reported some form of disability.

Anecdotal evidence of the positive effects of cannabidiol (CBD) oil on uncontrolled seizures has caused a stir in the epilepsy community and calls have been made to study its effects (Cilio et al. 2014). The growing literature available for this derivative of marijuana suggests a promising link between CBD oil and reduced seizure activity (Szaflarski and Bebin 2014; Gaston and Friedman 2016; O’Connell, Gloss, and Devinsk 2016). On 4/1/2014 a law was signed by the governor of Alabama allowing for a clinical study of a form of CBD oil to be conducted at the University of Alabama at Birmingham. The study commenced on 4/1/2015 and will ultimately examine approximately 140 children and adults longitudinally over three years. As part of the protocol, interviews are conducted with patients or their family members using structured questionnaires collecting social determinants of health (SDH) in epilepsy data at the initial enrollment visit then again at the 12-month mark. These data include social variables such as household income, sex, sexual orientation, social connections, race, ethnicity, perceived health, education level, health literacy, degree of religiosity, family make-up, financial strain, and stress inventories (Szaflarski 2014). In the first year of the study, 42 pediatric and 39 adult patients were admitted to the study and completed initial interviews. Patients were predominantly non-Hispanic white (96%) with 76% living in a home with an annual family income of at least $40,000 per year. Sex of the patients was pretty evenly split (51.2% female), the average age was 19 years, and few reported financial strains that included food scarcity or the inability to afford antiepileptic medication. The social profiles from this first-year study sample suggest that there are possible racial and socioeconomic barriers to accessing CBD treatment (Szaflarski et al. 2017).

Because of the high percentage of disability in this study group, most patients are unable to participate in interviews, and the social variable data are primarily provided by family caregivers. The structured nature of the interview questionnaire allows for consistency in response categories, also providing an opportunity for respondents to speak freely if they wish. Caregivers frequently give examples of the frustrations with family members who avoid them, stares and rude comments from passersby in grocery stores and restaurants, and the general lack of assistance in providing care for their child. Financial difficulties associated with obtaining medical equipment, lack of ability to work outside the home, and the exorbitant costs of epilepsy medications greatly impact the caregivers' abilities to afford respite care, replacement wheelchairs, or at times enough food to last through the end of each month. At times, some parents cry while answering questions about their children's social interactions, school activities, and friends. Some children have few or no friends because of cognitive delays or behavioral problems. For parents of the children profoundly intellectually disabled, these questions about sports teams and clubs are reminders that their children are not participating in normal childhood activities. One mother even called the questions "cruel."

The caregivers' expressed feelings of loneliness and hopelessness over their situations provided motivation to write a grant proposal for caregiver services in epilepsy. During the literature and data gathering phase of the grant-writing process, it became evident this was a neglected area of research. With the vast majority of the available data concerning caregiving concentrated on senile dementia and Alzheimer's disease, information documenting the needs and concerns of caregivers of PWE was noticeably absent. This dearth of literature put the grant proposal on hold and provided inspiration to conduct a study to examine the possible effects of perceived epilepsy stigma on the level of burden experienced by caregivers of patients with intractable epilepsy and to determine how this effect may vary with the age of the patient. In addition to providing data for this dissertation, comments gathered concerning practical needs of caregivers may be used to inform policies designed to provide services to caregivers of this patient population.

Dissertation Structure

This dissertation consists of six chapters, including Introduction (Chapter 1). Chapter 2 presents the academic literature on stigma and caregiver research. The first section of the chapter examines stigma research generally, then with an emphasis on epilepsy stigma. Topics include stereotypes of PWE, the similarities between mental health stigma and epilepsy stigma, and the "stigma coaching" (i.e. how to cope with stigma) PWE and their families receive at the time of diagnosis. The second section discusses caregiver burden research beginning with the studies of the challenges familial caregivers faced in the 1950s and 1960s following the mass exodus of mentally ill individuals from hospitals and asylums. Included are trends in defining and categorizing burden as the concept evolved into the multi-faceted phenomenon as it is currently understood. Finally, caregiver burden in epilepsy is also discussed across four domains, which include financial, physical, social, and psychological aspects of caring.

Chapter 3 outlines the theoretical frameworks of the study, describes conceptual approaches utilized in prior studies of stigma and caregiver burden in epilepsy, and introduces this study's conceptual model. Theoretical perspectives of stigma are presented. Caregiver burden is examined through the lens of the Stress Process Model (Pearlin 1991). The chapter then concludes with a description of this study's conceptual model and stated hypotheses.

Chapter 4 describes the study design and methodology. The study population and sample are identified, and the recruitment plan is presented. Variable operationalization, survey items, and coding labels are explained. Analytical strategies are outlined. Statistical results and tables are then presented in Chapter 5.

Chapter 6 discusses the study results, study limitations and implications for future research in the social experience of caregiving for PWE. Study conclusions round out the discussion.

CHAPTER 2

REVIEW OF LITERATURE

This chapter begins with an overview of what epilepsy is and how seizures have been perceived over time. It is not difficult to imagine that in early recorded history seizures were considered bad omens and that PWE were thought to be possessed by demons or evil. Over time, these perceptions have evolved into the current understanding that epilepsy is a neurological disorder. Despite this evolution this knowledge has not eradicated the stigma associated with seizures. Academic literature from the study of stigma is presented, followed by a section describing how stigma relates to epilepsy. A discussion of caregiver burden follows in much the same way. Brief definitions are provided to place the literature into context. The chapter ends with a summary of findings relevant to this study's conceptual model.

Epilepsy

Epilepsy Defined

Epilepsy, which is characterized as having two or more unprovoked seizures, is one of the most common chronic neurological disorders in the world, currently affecting approximately fifty million individuals (Shorvon 1996; WHO 2016). According to the World Health Organization Epilepsy Fact Sheet (2016), seizures are involuntary movements caused by excessive electrical discharges in the brain and involve either part of the body, aptly labeled “partial” or “focal”, or the entire body, labeled “generalized”. Seizures can include a loss of conscious and/or loss of bladder or bowel function. Treatment for epilepsy is effective in 70% of cases and typically involves one AED and then discontinuation of the drug after two years if the patient is free from seizures (Callaghan, Garrett, and Goggin 1988).

Types of Seizures

Depending on which part of the brain is producing the discharges, seizures can present with various symptoms. Considering the limited non-clinical scope of this paper, simplified seizure type definitions from the ILAE 2017 Classification of Seizure Types – Basic Version (Fisher, Shafer, and D’Souza 2016) are used. Under this newly adopted classification system, there are three basic features considered by neurologists when identifying what type of seizure a patient has: 1) location of seizure origination, 2) level of patient awareness during the seizure, and 3) other characteristics and features of the seizure.

First the location in the brain producing the abnormal electrical discharge is determined. If seizures originate in only one side of the brain, it is called a “focal” seizure. This term replaces the previously used term “partial.” Seizures that originate in both sides of the brain are known as “generalized” seizures. If seizures begin in one side and spread to both sides they are called “focal to bilateral” seizures (Fisher, Shafer, and D’Souza 2016).

How aware PWE are during a seizure is important to know, not only for classification purposes, but for safety reasons. During seizure events, even if unable to walk or talk, if PWE are aware of their surroundings, these are known as “focal aware” seizures. If awareness is impaired without a clear understanding of what happened, the seizure is classified as “focal impaired.” For persons who live alone, have seizures at night, or are unable to communicate awareness, their seizures are classified as “awareness unknown.” These awareness characteristics only apply to focal seizures. All generalized seizures are considered to have impaired awareness so they are not identified with a level of awareness in the label (Fisher, Shafer, and D’Souza 2016).

The final feature that determines how a seizure is classified is when considering other symptoms as they are related to body movement. A “focal motor” seizure occurs when the body jerks, stiffens, or twitches or automatisms occur, such as smacking lips, rubbing fingers or hands together, or running. When changes in emotions, thought processes, or sensations occur first, they are known as “focal non-motor” seizures. “Auras” are used to describe symptoms PWE might feel at the beginning of a seizure. For generalized seizures, when parts of the body are engaged, they are labeled as “generalized motor” seizures. Grand mal seizures are now classified using this label. If the generalized seizure does not affect body movement or the PWE stares into space or has repeated lip-smacking, the seizure is classified as “generalized non-motor” seizure. These seizures were formerly known as petit-mal (Fisher, Shafer, and D’Souza 2016).

Intractable epilepsy

Approximately 30% of PWE have intractable epilepsy. Epilepsy is considered intractable or refractory when seizures are not relieved by medications or other treatments, typically after two failed AED medication failures or problems with tolerating the drugs

(Kwan and Brodie 2000). Other treatments for epilepsy besides medication might include brain surgery, special diets, vagus nerve stimulation, electroshock treatments, or natural remedies such as vitamins, herbal supplements, and exercise (Wheless 2006).

Intractable epilepsy adversely affects the physical and mental health of PWE, both children and adults, and interferes with their normal social functioning (Devinsky 1999). Health-related quality of life (QOL) decreases as the number of seizures increase for PWE with psychological and social domains being particularly problematic (Leidy et al. 1999). Wheless (2006) conducted a study of PWE examining how refractory seizures affected the lives of patients. More than 50% of respondents reported physical and psychological effects of seizures and medications that included difficulty thinking clearly, memory loss, drowsiness, lethargy, clumsiness, muscle twitches, and depression. Around half of all study respondents wanted to be more socially active but felt their epilepsy held them back from participating. Feeling independent was a desire of most of the PWE surveyed. Many felt their poorly-controlled seizures made them a burden on their loved ones and nearly 50% reported they chose to drive rather than ask others for a ride. For the group able to speak for themselves, education rates were high at 64% graduating from high school and 26% having at least a bachelor’s degree. Despite being highly educated, this group had an unemployment rate of 29%. Those with caregivers answering for them reported that epilepsy substantially limited patients’ abilities to participate in activities of daily life.

Stigma

Stigma Defined

For thousands of years PWE have been perceived as deviant; in pre-modern cultures they were thought to be demonically possessed or marked by God and their seizures considered as bad omens (Temkin 1994). Often associated with mental illness or derangement, PWE were, and still are by some, believed to be evil, dangerous, sexually deviant, aggressive and unreliable (Prioreschi 1996). Although public attitudes concerning epilepsy have changed for the better, there are still instances of prejudice and discrimination at the interpersonal and institutional levels (Jacoby 2002; Jacoby and Austin 2007).

Prejudice is an attitude towards someone because they belong to a particular group while discrimination is the act of treating that person differently (Thornicroft et al. 2007). In epilepsy, prejudice and discrimination often stems from stigma (Jacoby 2002; Elafros et al. 2013). Technically, the definition of stigma is a mark or brand on the body to signal to others that the person is tainted and is to be shunned. Over time, the meaning of stigma has changed to include any characteristic, whether a physical mark or a character attribute, that is associated with shame. It is, in effect, a relationship between an attribute and how that attribute is symbolized by others (Curra 2014).

Stigma Components

Stigma components can be sorted into categories determined by considering from whom the stigma is imputed or by whom the stigma is perceived. Public stigma, also known as enacted stigma, focuses on attitudes of a general population toward someone who is different (Goffman 1963; Link and Phelan 2001). The flip-side of enacted stigma is perceived or self-stigma, where the stigmatized person internalizes society's negative views of them and feels shame for their condition resulting in attempts to hide the condition if possible (Goffman 1963; Link and Phelan 2001; Scambler 2009). Family stigma is a collection of discriminatory actions and attitudes experienced by relatives of a stigmatized person, enacted by people in the general public. In other words, this kind of stigma can be described as courtesy stigma or a "stigma by family association" (Goffman 1963). Mak and Cheung (2008), in looking at family caregivers of stigmatized persons, differentiated between perceiving stigma by family association and the internalization of those societal views. This internalization coupled with the psychological responses to the stigma results in affiliate stigma, which this study incorporates into its conceptual model.

Stigma in Epilepsy

It is important to understand the stigma associated with epilepsy in order to grasp the stigma caregivers of PWE might perceive. Characteristics of epilepsy stigma mirror mental illness stigma in several ways. Stigma involves negative stereotypes and some of the characteristics associated with mental illness are: passive, ignorant, worthless, sick, foolish, unpredictable, and dangerous or prone to violence (Tausig, Michello, and Subedi 2004; Schnittker 2013). Rogers and Pilgrim (2014) argue that there are three qualities a mentally ill person possesses that lead to stigmatization: 1) a lack of intelligibility, or an inability to explain one's own behavior, 2) lack of social competence, where the person is unable to function in a normal social role, and 3) the presence or threat of violence. The problem with using these qualities when describing a mentally ill person, or a person with epilepsy for that matter, is that a person may seem unintelligible at times, but this is rarely a constant state (Pilgrim 2009). Regardless, once a person is made aware of another's mental illness, they may react with fear, hostility, disgust, and/or contempt. Reactions typically result in the tarnished, or stigmatized, person being separated from "normal" people and they suffer consequences caused by the social distance (Pilgrim 2009; Schnittker 2013). The stigma experienced by family members of those with mental illness diagnoses occurs across several domains, including enacted and perceived, at interpersonal and institutional levels (Muhlbauer 2002).

The negative characteristics associated with mental illness are also reportedly associated with epilepsy. At the interpersonal level people still acknowledge that PWE are treated differently, avoided, and excluded from social activities (Dilorio et al. 2004). Negative stereotypes about epilepsy include the ideas that PWE are unreliable, should not marry or have children, are dangerous, unable to maintain work responsibilities, and unable to function in social roles. Avoidance of PWE seems to be associated with fears of epilepsy being contagious, not knowing what to do during a seizure, or the belief that the PWE was likely to die during a seizure (Jacoby et al. 2004). Although in many countries PWE fall into a protected class with legal protection, discrimination at the institutional level still occurs when it comes to employment. For instance, in the U.K., those with epilepsy are not allowed to join the armed forces and there are restrictions if they become teachers, doctors, police officers, or fire fighters (Jacoby and Austin 2007).

Stigmatization is a learned behavior and stigma in health conditions like epilepsy is no different (Scambler 2009). From the first moment of diagnosis, a PWE perceives stigma from both doctors and well-intentioned family members. The expectation of enacted stigma drives the “stigma coaching” they receive, which is usually peppered with advice on how to deal with the comments, looks, and discrimination from others as well as ways to hide their epilepsy (Schneider and Conrad 1980).

Interestingly, the reality of high levels of enacted epilepsy stigma may be overestimated. Scambler and Hopkins (1986) challenged the assumption of overt discrimination practices toward PWE by offering up an alternative viewpoint to the orthodox viewpoint of epilepsy stigma. The orthodox viewpoint incorporates four underlying beliefs: 1) there is ignorance on behalf of the public about the true nature of epilepsy, 2) the public in general has negative attitudes toward PWE, 3) discrimination is to be expected by this ignorant public, and 4) discrimination perpetrated by the ignorant public is to blame for the problems related to the epileptic identity.

Scambler and Hopkins further noted that results from qualitative studies in 1983 and 1986 indicated that there is little evidence to support any of the four components of the orthodox viewpoint. They found that PWE experience felt stigma before experiencing enacted epilepsy stigma and that their expectation of stigma originated with the stigma coaching they received. In an alternative model, the researchers suggested that epilepsy stigma is derived from a fear of enacted stigma and that, because it is communicated to them upon their diagnoses, they learn that their new status as a PWE is socially undesirable. The second component of this alternative model is that the predisposition to hide their status as a PWE is also derived from fear of enacted stigma. A final component is that felt stigma is more likely to disrupt the life of a PWE than enacted stigma.

The orthodox viewpoint is such a given that it has not only shaped the public perception of epilepsy stigma, but has informed policies addressing how medical providers counsel PWE about dealing with their diagnoses of epilepsy including handling inevitable stigma and discrimination (Schneider and Conrad 1980; Scambler and Hopkins 1986). In fact, there is evidence that when self-reporting, epilepsy is underreported in certain patient populations because of the relabeling of epilepsy as "seizure disorder" upon conferral of the diagnosis. In a population survey in Washington, DC, 75% of respondents with a history of epilepsy identified themselves as having a seizure disorder rather than epilepsy with many not understanding they were considered to be the same. This tendency was most associated with females, non-whites, those older than 50 years, having a high school education or less, and low income (Kroner et al. 2016).

Parents of pediatric epilepsy patients may be more affected by others' negative views toward epilepsy than their children are and may be more likely to perceive epilepsy stigma (VanStraten and Ng 2012). Jacoby and Austin (2007) suggest that pediatric patients' attitudes toward epilepsy begin with how their parents react to the diagnosis. When parents believe that epilepsy results in discrimination or hostile reactions, their children learn that epilepsy is shameful and consequently will seek ways to hide their condition. With teachers and medical providers echoing this mindset, children learn to expect discrimination and internalize the feelings of being less favorably regarded leading to lower self-esteem and increased feelings of depression and anxiety (Collings 1990). This felt stigma seems to have a positive association with seizure frequency (Jacoby 2002), and those with intractable epilepsy might experience high levels of felt stigma.

Familial caregivers of PWE are also susceptible to internalizing the stigma of epilepsy resulting in affiliate stigma. As Mak and Cheung (2008) explain, affiliate stigma is not only the internalization of stigma by the family member but also includes their resulting psychological responses. Affiliate stigma has been associated with increased anxiety, depression, and stress (Rodenburg et al. 2007; Mak and Cheung 2012). As with other health-related stigmas, higher levels of education and knowledge about epilepsy are associated with lower levels of affiliate stigma while less perceived community and family supports were associated with more affiliate stigma (Elafros et al. 2013). Affiliate stigma in epilepsy has not been studied in this manner and is typically associated with caregiver or patient QOL and almost exclusively in relation to caring for a family member with autism, a developmental disability, or a mental illness (Mak and Cheung 2008; Mak and Kwok 2010; Mak and Cheung 2012; Chiu et al. 2013; Werner and Shulman 2013). Further research needs to be conducted to provide a clearer picture of how this type of stigma intersects with health outcomes of family members, as well as how affiliate stigma is associated with physical health problems such as epilepsy.

Caregiver Burden

Caregiver Burden Defined

Providing care for a family member with intractable epilepsy can impose a tremendous financial, physical, social, and psychological burden on a caregiver, possibly because of the unstructured and unsupported nature of the arrangement (Karakis et al. 2014; Smith et al. 2014). Unpaid informal caregiving occurs between a patient and someone who has a preexisting personal relationship with each other, usually between family members (Chou 2000).

Caregiver Burden in Epilepsy

Researchers use various indicators to measure burden, such as indices that examine the QOL for caregivers, caregiver stress inventories, child behavior checklists, psychological symptoms checklists, and some use stigma results as a proxy for caregiver burden or incorporate stigma as a type of burden (Austin et al. 1998; Farrace et al. 2013; Karakis et al. 2014). Most of these measures focus on stress, anxiety, and psychological reactions to caregiving and use the total scores in quantitative analyses as either predictors or outcomes. This can be problematic when one takes into consideration the broader understanding of caregiving burden. Qualitative interviewing and focus groups to identify thematic aspects of burden have been found to be effective in identifying burden themes but are typically small in sample size and difficult to generalize to a broader population (Collings 1990; Wagner et al. 2009).

The aspects of caregiving in epilepsy closely mirror the characteristics of caregiving in other chronic diseases. Caregivers experience prolonged stress over years, sometimes with increasing intensity and find themselves with fewer and fewer social connections, particularly when their family member has intractable epilepsy (Smith et al. 2014). The expenses associated with caring for a PWE, particularly someone with intractable epilepsy, are not only assessed with direct costs for wheelchairs, durable medical supplies such as helmets, walkers, incontinence supplies, or medications, but also with reduced family income (Gibson 2014). Due to the need for round-the-clock care and lack of resources, the decision is frequently made for at least one family member to quit working in order to stay home and provide that care (Smith et al. 2014).

Acting as primary caregiver for PWE can be detrimental to one's overall health and, not surprisingly, those caring for family members with intractable epilepsy report both reduced well-being and high levels of caregiver burden (Van Andel, et al. 2009; Farrace et al. 2013; Gibson 2014; Karakis et al. 2014). Physical health of caregivers often suffers because of stress from worrying, the physical demands of lifting, carrying, bathing, and other caregiving activities, and lack of sleep. Sleep issues, which include sleeping lightly, sleeping fewer hours, or co-sleeping with the PWE, contribute to the overall burden (Gibson 2014; Smith et al. 2014). Stress and anxiety levels are particularly high for parents of children with intractable epilepsy (Wirrell et al. 2008; Farrace et al. 2013).

Psychosocial stressors for caregivers may include increased family strife, the need to constantly advocate for the family member, problematic behaviors of the PWE, social isolation, the need to dedicate a great deal of time to caregiving activities, changes in caregiver career trajectories in order to accommodate the family member's needs, and changes in marital and family dynamics (Farrace et al. 2013; Gibson 2014; Smith et al. 2014; Thompson et al. 2014; Hamama-Raz and Hamama 2015). Because of the unpredictable nature of intractable epilepsy, caregivers frequently report they must maintain constant vigilance and worry incessantly about seizure triggers (Buelow et al. 2006; Gibson 2014). The disruption of day-to-day life can be overwhelming for families, especially when epilepsy is first diagnosed. Planning outings as a family becomes nearly impossible and families report being split with one member staying home to care for the PWE while others attend church, shop, work, or otherwise engage in activities (Buelow et al. 2006; Gibson 2014; Thompson et al. 2014).

Despite the challenges of caring for a family member with intractable epilepsy, up to one-third of caregivers report their families become closer and more resilient and they themselves experience a new-found purpose in educating the public and advocating on behalf of their family member (Smith et al. 2014; Thompson et al. 2014). These caregivers appear to have adequate coping mechanisms to deal with stressors. Among the positive aspects of caregiving in epilepsy are contentment with life, resiliency, high levels of family cohesion, lower levels of stress, and a strong social support network through churches and other parents with disabled children (Buelow et al. 2006; Rodenburg et al. 2007).

The majority of studies of familial caregivers in epilepsy focus on those caring for children with very few examining family members taking care of adult relatives. In two such studies that examined family caregivers of adult patients, results were similar in that older caregivers reported lower levels of stress and higher QOL scores. Burden levels were negatively associated with the number of AED medications the patient was on and cognitive performance rather than specific caregiving activities (Llewellyn et al. 2010; Karakis et al. 2014).

Of the literature reviewed for this chapter there were no studies examining stigma and caregiver burden as separate concepts within the same study. The research typically examined epilepsy stigma as it was perceived by the adolescent or adult PWE rather than the familial caregiver. The studies that did focus on caregivers typically looked at parents of children and how the parents’ perception of stigma translated into increased senses of stigma by the child. Stigma, occasionally, was measured as a component of overall burden, but was not independently measured or differentiated, nor was it considered as a possible predictor for caregiver burden levels. As well, there were no studies identified that examined burden experienced by caregivers of both pediatric and adult populations, and there were very few focusing on caregivers of adults. Because there are so few studies examining the burden associated with caring for adult PWE, this study is of particular importance. Filling this gap will provide a broader understanding of the lived experience of caring for family members with intractable epilepsy.

CHAPTER 3

THEORETICAL FRAMEWORK, CONCEPTUAL MODEL AND HYPOTHESES

In this chapter, the theoretical framework for this study is discussed. The evolution of stigma theory is traced and trends in stigma research are outlined. The concept of caregiver burden is situated within Pearlin’s (1991) Stress Process Model before discussing trends in burden research. Previous theoretical approaches to the study of stigma and caregiver burden in epilepsy are presented to frame the need for this approach. The conceptual model for the study is introduced and diagrams of the conceptual models are included to provide a visual reinforcement of the model com-ponents. The chapter ends with specific study hypotheses stated.

Theoretical Frameworks of Stigma

The sociological study of stigma can be traced back to Erving Goffman (1959) when he explained how individuals present a façade to fit in to each situation, in effect playing roles as if in a play, with a goal of achieving normality. According to Goffman (1959; 1963) when a person falls outside the framework of normal, that is, between what a person is and what a person should be, that person becomes deviant. A deviant person is stigmatized by others and one of the most challenging things about dealing with a physically apparent stigma is trying to hide it using impression management techniques. Goffman presented the idea that there are three types of stigma: tribal (characteristics belonging to a group), discreditable (a blemish of character), and discredited (an abomination of the body) (1959 Pp. 4-5).

Link and Phelan (2001) expanded Goffman's concept of stigma and incorporated five components: 1) the labeling of human differences, 2) the linking of those differences to negative stereotypes using dominant cultural beliefs, 3) placing those who are different into categories designed to separate them from the "normal" group, 4) unequal outcomes for those that are different because of loss of status and discrimination, and 5) a power construct put into place that allows all of the labeling, separation, and discrimination occurring. The stigmatized person's loss of status and his/her perception of being discriminated against distinguish this theory from Goffman's. These two factors affect life chances in many areas, including mental and physical health.

More recently, Pescosolido and Martin (2015) further distinguished between the different categories, incorporating them into one overarching theoretical framework. These five components include: 1) perceived stigma where there is agreement that discrimination and prejudice against a labeled group exists, 2) anticipated stigma where someone in that labeled group expects to be devalued and discriminated against, 3) endorsed stigma where there is agreement with the stereotypes associated with the stigmatizing feature, 4) received stigma which focuses on the stigma from the perspective of the person personally experiencing discrimination, and 5) enacted stigma where individuals discriminate against or act prejudicially toward a stigmatized person.

Trends in Stigma Research

Previous theory work has led to current trends in stigma research. Three areas of focus in current stigma research are examined here. It has been suggested that researchers should look at stigma as a moral process, where a person's local social world dictates the norms for them. If they cannot operate within those norms, they are stigmatized. Kleinman and Hall-Clifford (2009) noted that stigma has a social distribution shaped by social, economic, and political power. In any local environment, wealth, relationships, and life chances matter greatly to individuals and stigma may make it difficult or impossible for a person to participate fully, thus presenting a moral dilemma. They suggested that researchers conduct ethnographic studies and combine qualitative with quantitative data to inform policies to reduce the negative consequences of stigma.

A second focus of stigma research attempts to answer the question, "Why do people stigmatize others?" Goffman (1974) suggested that people stigmatize others to enact social control on others. People are uncomfortable when others appear different than expected and that strain they feel needs to be addressed. The easiest way is to act in such a way that motivates the deviant person to step back in line. Stigma is based on a negotiated understanding of social roles and expectations, which are not universal (Curra 2014). Phelan, Link, and Dovidio (2008) suggested that people stigmatize others as a means to three ends: the goal of keeping people down through exploitation and domination, the goal of keeping people in through enforcement of social norms, and the goal of keeping people away to promote the avoidance of disease, which often occurs with health-related stigmas.

A third trend in current stigma research involves examining stigma power (Link and Phelan 2014). Considering the idea that people stigmatize others to get something they want, whether it is keeping people down, in, or away, it is an easy leap to understanding that accomplishing this takes power. In this research trend, the primary focus is on determining what people have to gain by imputing stigma on others and how they wield stigma power. Based in Bourdieu's concept of symbolic power (Bourdieu, 1985; Swartz 1997), those with power are able to impose their versions of what is of value, thus establishing the norms for a group. Link and Phelan found three takeaways from Bourdieu applicable to their concept of wielding stigma power. First, the mechanism through which power is exercised is what a culture deems valuable and stigma is an indicator of what has value. Second, when a stigma has been internalized, even the person who is stigmatized accepts it, even if it harms them. Third, symbolic power is frequently woven into society so intrinsically that it is a taken for granted aspect of culture, in essence it is "misrecognized".

Stress Process Model

Caregiver burden revolves around the theoretical frame of the stress process and coping. The Stress Process Model has four domains: 1) the context of the stress, 2) the stressors, 3) the mediators, and 4) the outcomes of the stress (Pearlin et al. 1981). Stress is a byproduct of social demands and/or the absence of means to attain what one needs and coping is a response to a stressor that prevents harm caused by that stressor. A primary stressor, such as caring for a chronically ill family member, can lead to secondary stressors such as financial loss, marital and family conflicts, or social isolation. Stress proliferation occurs when chronic stressors accumulate and, over time, may have a deleterious effect on overall health (Pearlin, Anashensel, and LeBlanc 1997).

Mediating resources most frequently considered in the Stress Process Model include coping, social support, mastery, and belief systems and values (Pearlin 1991). Coping actions are responses to stressors. These actions include behaviors the stressed person performs in an attempt to manage either the situation itself that gave rise to the stress, the meaning of the stressful situation, or the resulting symptoms of the stress (Pearlin and Anashensel 1986). Mastery is understood as individuals' beliefs in their ability to control what happens to them while belief systems and values act separately from coping and social support as mediators to stressors (Pearlin 1991). Interestingly, in the stress process model, mediators can be tricky in that they can sometimes also be moderators within the same model (Pearlin and Bierman 2013). For example, mastery serves both functions. Prolonged economic strain on a caregiver may reduce levels of perceived mastery, which in turn, increases depression levels. At the same time though, higher levels of mastery tend to weaken the relationship between financial strain and depression.

Caregiving burden in chronic illness is a phenomenon that seems to perfectly exemplify stress proliferation. When Pearlin and his colleagues (1990) examined caregiving through the stress process lens, they differentiated between "caring," the affective aspect of committing to the welfare of someone with whom a caregiver has an established role, and "giving," which is a behavioral aspect within that role. When someone in a family has a lingering or progressive impairment, the caregiving role evolves as the ill family member's needs grow and eventually becomes the primary role. Where the relationship role might once have been wife/husband or mother/child, it becomes caregiver/care-recipient and becomes all-encompassing imposing on other roles the caregiver might have performed before.

Burden perception changes over time as primary roles evolve. In the cases where a disease or condition lingers or gets progressively more debilitating, the informal caregiver becomes more involved with direct caring activities, which increases the possibility of additional stressors. Coping actions that worked before may not meet the caregiver's need to manage the proliferated stress and must change with the new situation (Perlick, Clarkin, and Sirey 1995). Also the relationship rewards may become one-sided with the care-recipient receiving the majority of benefit from the relationship and the caregiver receiving fewer benefits as time goes on and feeling overwhelmed or overloaded (Corbin and Strauss 1988; Pearlin et al. 1990). At the same time, some caregivers report an acceptance of their situations and stress becomes less impactful on their health (Pearlin et al. 1990; Perlick, Clarkin, and Sirey 1995).

Trends in Caregiver Burden Research

In early caregiver burden research, the focus was on how to conceptualize burden. These conceptualizations were rooted in academic literature published during the transition of mentally ill individuals from institutionalized care to community based care in the 1950s and 1960s. Hoenig and Hamilton (1966), in an effort to differentiate between the types of burden experienced by caregivers, divided burden into two categories: objective and subjective. Objective burden referred to the negative, observable, and measurable impact of caregiving while subjective burden referred to the unpleasant feelings caregivers may have about their caregiving activities. Financial costs, changes in employment and income, separation of a parent from a child due to caring for the relative, distress felt by other members of the household, decreased health status of the caregiver or other household members, and any disruption to routine daily activities were categorized as objective burden. Interestingly, disturbing behaviors on the part of patients with schizophrenia were grouped under this category as the behaviors were observable and were thought to interfere with the caregiver's health. General feelings of "no burden," "some burden," or "severe burden" were categorized under subjective burden and were vague in operationalization. If the caregivers reported feeling severely burdened, rather than categorizing it as subjective burden, it was usually rationalized that the burden was in relation to the odd behaviors their family member with schizophrenia were exhibiting and was ultimately categorized as objective in nature.

The notion of evaluating what in particular was considered burdensome about providing care for a family member was introduced in a 1966 British study of families caring for mentally ill relatives discharged from hospital and institutional settings to homes with ancillary services in place (Grad and Sainsbury 1966). The researchers' examination of effects on the caregiving families led to an operationalization of the concept of family burden, measured by determining a "problem score" based on how affected families were by changes in income and employment, ability to participate in social and leisure activities, the health of other family members in the home, and relationships with neighbors. This was one of the first times the idea that decreases in social and leisure time were considered to be aspects of caregiving burden. Results indicated that families with more services and relief from stressors such as day programs, home visits, and access to nursing home care had lower burden scores.

As caregiving burden was increasingly examined, the definitions of objective and subjective burden became more refined. By the mid-1980s burden was still measured as a dichotomous variable; objective burden became understood as measurable financial and physical costs of caregiving while subjective burden described personal feelings expressed by and the emotional toll taken on the caregiver (Platt 1985). The idea that the burden of caregiving was negative due to the activities involved had evolved into an understanding that burden was a process (Zarit, Reever, and Bach-Peterson 1980).

Currently, the conceptual understanding of caregiver burden encapsulates any and all problems family members experience associated with providing care for another family member with an impairment or chronic illness (Chou 2000). Caregiver burden is a multi-faceted phenomenon with at least four burden domains: financial, physical, social, and psychological (Woods, Haberman, and Packard 1993; Chou 2000). Financial costs include money spent on supplies and increased daily living and medical expenses, transportation to and from doctor and therapy appointments, and durable medical equipment. The physical costs include the wear and tear on the caregiver's body from lifting, bathing, dressing, or changing their family member. Physical toll also includes compromised physical health due to anxiety, depression, lack of sleep, constant stress, and injuries sustained from combative patients. Social isolation from family, friends, and previous colleagues is an aspect of caregiving as well. Psychological stress from worry, emotional fatigue, loss of career goals, and the demands made by their family member contribute to overall burden.

Conceptual Model

The conceptual model for this study was developed after reviewing academic literature on stigma and caregiver burden in epilepsy. Anecdotal evidence from caregivers completing SDH interviews during their family members' enrollment into the CBD study at the University of Alabama at Birmingham reinforced the need to consider stigma separately from caregiver burden. While some family members spoke of high levels of perceived stigma or burden levels, others said they had not noticed discrimination of their family member at all. Burden seemed to be a separate concern yet stigma was frequently considered as part of caregiver burden in epilepsy in other studies.

When observing stigma through the lens of Pescosolido and Martin's (2015) stigma framework, there is evidence that all five components apply to intractable epilepsy. First, there is agreement that there is a perception of discrimination and prejudice occuring against people with intractable epilepsy (Scambler and Hopkins 1986). Second, it is anticipated that PWE will be discriminated against as evidenced by stigma coaching received by patients and their families (Schneider and Conrad 1980; Jacoby and Austin 2007). Third, there is endorsement by PWE of some of the stereotypes about epilepsy, mainly that they are unpredictable, can be a burden on others, and have issues with employment (Wheless 2006). Next, there is a perception that the discrimination a PWE is receiving is because of epilepsy (Scambler and Hopkins 1986; Jacoby and Austin 2007; VanStraten and Ng 2012). Finally, there is evidence of enacted epilepsy through instances of discrimination (Dilorio et al. 2004; Jacoby and Austin 2007).

The decision was made to conceptualize perceived stigma in this study using Mak and Cheung's (2008) frame of affiliate stigma. If this frame is correct affiliate stigma occurs when caregivers internalize the stigma they perceive is directed toward them and their family members and have psychological responses to it. In this study, caregiver burden might be interpreted as a psychological response in affiliate stigma as the instrument being used to gather data indicates how stressful each caregiving item is to the respondent.

A positive relationship was expected between levels of perceived epilepsy stigma felt by caregivers and the levels of burden those caregivers perceived (Figure 1). The higher the level of stigma experienced by a caregiver, the higher the level of perceived burden. Additionally, it was hypothesized that age might mediate the relationship between perceived epilepsy stigma and perceived caregiver burden (Figure 2). During the course of SDH interviews in the CBD study, while most patients' families reported challenges in caring, parents of younger patients reported little or no discrimination while familial caregivers of older family members reported discrimination more frequently. It was also hypothesized that age moderates this relationship -- in that the older the PWE was, the stronger the relationship between perceived stigma and perceived burden experienced by the caregiver (Figure 3). This possibility stemmed from observed differences between parents in the two groups. Parents in the adult patient group seemed more resigned and more tired; several of the parents who commented on their feelings of isolation said they had lost most of their friends over the years because their friends had become nervous around their children or were impatient that they were unavailable for social gatherings.

Figure 1. Conceptual Model, Association Between Stigma and Caregiver Burden

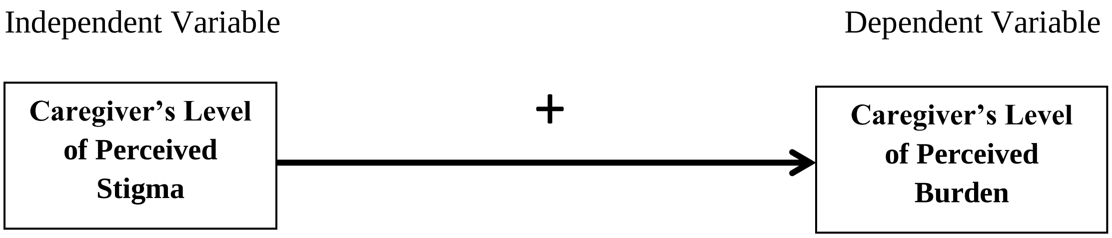


Figure 2. Conceptual Model, Age as Mediator

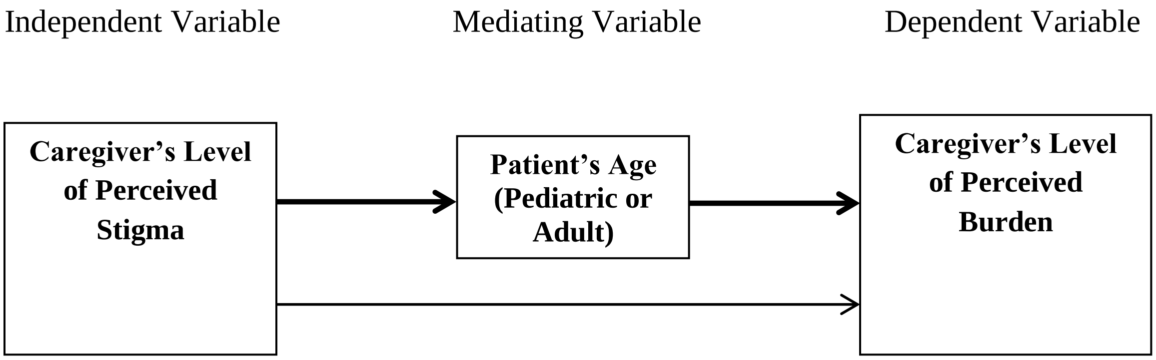
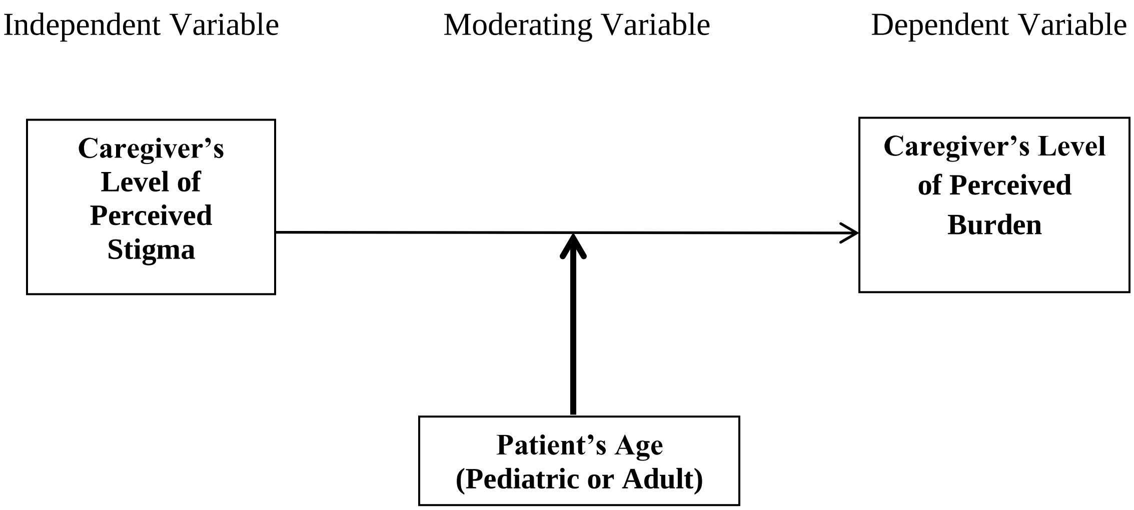


Figure 3. Conceptual Model, Age as Moderator



Hypotheses

The conceptual models yielded the following hypotheses:

Hypothesis 1:

H_1: There is a positive relationship between perceived epilepsy stigma and perceived burden among caregivers of individuals with intractable epilepsy. As a caregiver's level of perceived stigma increases, their perceived level of burden increases.

Hypothesis 2:

H_2: The relationship between perceived epilepsy stigma and perceived burden is mediated by the age of the family member with epilepsy.

Hypothesis 3:

H_3: The relationship between perceived epilepsy stigma and perceived burden is moderated by the age of the family member with epilepsy.

CHAPTER 4

RESEARCH DESIGN AND METHODOLOGY

This chapter presents the research design and methodologies used in this study. First, the study population and eligibility for participation are outlined. Next, the study design is described with data collection strategies, and the recruitment plan and sample are identified. The measurement and analysis sections follow.

Study Population and Eligibility to Participate

The study population for this study was comprised of family caregivers for individuals diagnosed with intractable epilepsy. A confirmed diagnosis of intractable or refractory epilepsy was necessary for inclusion in the study. In order to be diagnosed with intractable epilepsy, patients must have recurrent seizures and a history of two or more AED medication attempts that either fail to control seizures or produce intolerable side effects (Devinsky 1999; Kwan and Brodie 2000). Due to the confirmed diagnosis requirement, only caregivers of PWE being treated for epilepsy by a neurologist were accepted into the study.

The most common test to confirm the diagnosis of epilepsy is an electroencephalogram (EEG) or a video electroencephalogram (VEEG) (Shorvin 1996; Schacter, Shafer, and Sirven 2013). Depending on seizure frequency, a patient may be monitored for a few hours or be admitted to the hospital for several days. During this time, leads from a monitoring device are attached to the scalp and covered with a cap. Brain waves are recorded and attempts are made to provoke seizures using flashing lights, exercise, or sleep deprivation, as seizure triggers vary with each individual. This test helps determine from which part of the brain the seizures are originating, how frequent the seizures are and how long they last. Audio and video recordings provide valuable details for neurologists to examine when determining a course of treatment (Schachter, Shafer, and Sirven 2013).

Study Design

Data Collection

The cross-sectional and quantitative design of the study was supplemented with a few qualitative-style questions where respondents were allowed to list or describe answers freely. Data were collected using an original self-administered survey instrument (Appendix A). Demographic questions about the PWE for whom the family member provided care included age, gender, and whether they were enrolled in the Alabama CBD study. Demographic questions about the caregivers included age, gender, race/ethnicity, marital status, education level, income level, and number of adults and children in their home. They were also asked about how they heard about the survey. Measures of perceived caregiver burden and perceived affiliate stigma were included, as was an open-ended question about what services the family might find helpful. There was also an opportunity to comment on anything they would like at the end of the survey. These last two items were qualitative in style with the rest of the items quantitatively measured.

In order to determine inclusion in the study, the first question asked “Are you a primary caregiver to a family member with intractable epilepsy? (Check one)” with a possible yes or no response. For those answering “yes”, their data were included in the study. The second question asked “Is your family member enrolled in the Alabama CBD study? (Check one).” Three possible responses included “yes”, “no”, and “they were enrolled but are no longer in the study.” This question was included in order to provide an overview of perceived stigma and caregiver burden to the principal investigators of the CBD study for their general information. Study ID numbers were assigned to each completed survey. Per the IRB-approved protocol, signed consent forms (Appendix B) were stored separately from the surveys as an additional measure to ensure confidentiality for respondents. Consent forms were required to have a witness signature in addition to the respondent signature. Any forms without both signatures were excluded from the study.

Recruitment Plan and Compensation

This dissertation proposal was successfully defended on 4/7/2016 and, after complying with the Institutional Review Board (IRB) requests for changes in the protocol, final IRB approval was granted on 6/6/2016, protocol number 160401005 (Appendix C). The two principal investigators for the Alabama CBD Study gave permission to offer the survey to that study’s parental caregivers. CBD study patients were not approached at any time in order to not break that study’s IRB protocol. After permission was granted by treating physicians, caregivers of PWE treated in the general epilepsy pediatric and adult clinics were asked if they might wish to participate in the study. A flyer was created to post in the Epilepsy Monitoring Unit office to reach possible participants (Appendix D). There was no cost to participate and no compensation was offered for this research study.

Sample

The estimated sample size was predicted to range from 100-200 participants. All participants included in the sample were caregivers of a family member with intractable epilepsy regardless of enrollment in the CBD study. To determine the minimum sample size required for this study's analytical strategy, a power analysis was conducted using a formula from a website from California State University (Soper 2016). Data used in the formula included: significance level at $p < .05$, statistical power of .80 with a medium effect size ($f^2 = 1.5$). With two main predictors (stigma level and age category) this study needed a sample size of at least 67. With 3 predictors (stigma level, age category, and interaction term) there needed to be at least 76 respondents. Only family caregivers older than 18 years of age were allowed to participate in the study.

Data collection began on 6/23/2016 and ended 10/31/2016. Of 154 returned surveys, 136 were included in analyses. Three were returned without completed signatures on the informed consent. Fourteen surveys were incomplete. One survey was complete but the respondent indicated he was not a primary caregiver of a family member with intractable epilepsy and was excluded.

Measures

Dependent Variable - Caregiver Burden

The respondents' levels of caregiver burden were measured using the Carers' Assessment of Difficulties Index (CADI), which Nolan and Grant (1989) created to measure caregivers perceptions of the difficulties associated with caregiving. Used throughout Europe, this instrument was developed in England in response to that country's community care legislations in the late 1980s aimed at addressing challenges experienced by caregivers. Up until that time, there were very few assessments of family caregivers that were well-validated or relevant to diverse family circumstances. The CADI was one of three assessments designed to create a more complete picture of the challenges experienced by caregivers and packaged into a handbook for social workers (Nolan, Grant, and Keady 1998). In addition to the CADI, the Carers' Assessment of Satisfactions Index (CASI) assessed the satisfaction caregivers experience from providing care and the Carers' Assessment of Managing Index (CAMI) assessed coping strategies used by caregivers to manage stress. As explained in the handbook, a trained assessor first established trust and formed a therapeutic relationship with the caregiver. After completing the three assessments, the caregiver then worked with the professional to identify issues and help create a care plan to best meet their needs.

Dr. Mike Nolan, the primary developer of the instruments, was contacted via email, and the appropriateness of using the instruments for this study was discussed. He agreed with the request to use one of the instruments and provided a scanned copy of the original handbook, *Assessing the Needs of Family Carers: A Guide for Practitioners (1998),* as this book was out of print and could no longer be purchased. Because of the absence of therapeutic relationships with the respondents, only the CADI instrument was used as a means of identifying the most common difficulties, to obtain burden index scores, and to determine possible variations within sub-groups of caregivers. The greatest advantage of using the CADI was that it was appropriate for caregivers of all ages of impaired family members. For use in this study three modifications were made to the index to reflect American spelling and vernacular. The term "carer" was replaced with "caregiver”, “behaviour” was spelled “behavior”, and the phrase “plays me up” was replaced with “manipulates me” for clarity.

Another advantage was that, as a whole, the CADI has high internal consistency, $\alpha = .92$ (Nolan, Grant, and Keady 1998). The CADI, a unidimensional index measure, was intended for use as an assessment of individual circumstances of caregiving. These thirty individual items were combined into an index to measure overall scores of burden and could be divided into sub-scales or individual items when a caregivers’ needs were being considered and social care plans drawn up to provide assistance to families. Nolan and Grant (1992) conducted a principal components analysis (PCA) with varimax rotation on the thirty items that were being measured in order to create subscales within the index. If items had a factor loading of .35 or higher, they were included in the subscale and if an item loaded on to more than one subscale, it was only included in the highest loading one. At that time, seven subscales were identified including carer-dependent relationships, reactions to caregiving, physical demands of caring, restricted social life, poor family support, poor professional support, and financial consequences.

Two other PCA analyses have been conducted on the CADI instrument. Charlesworth and colleagues (2007), with a sample of 232 caregivers of adults with dementia in the U.K., conducted a PCA from which eight factors with eigenvalues greater than 1 emerged that explained 59% of the variance among burden levels. The eight components, in descending order, included: carer’s reaction to caring, degree of physical help, carer/person with disability relationship, restrictions on social life, professional support, family support, interpersonal demands, and financial consequences. This analysis differed from the original in that the researchers opted to score the CADI differently. Whereas

Nolan and Grant scored the response categories by both frequency and a level of stress experienced, Charlesworth's group combined them into a single rating. In doing so, however, several items that result from stress such as feeling helpless or having poor emotional well-being were loaded under the "carer's reaction to caring" component rather than a more specific component regarding those stressors (McKee et al. 2009).

Another PCA was conducted on the CADI (McKee et al 2009), this time scoring the CADI as Nolan and Grant (1992) had done. The study was conducted in the U.K., Italy, and Poland among 295 informal caregivers. Once the components were extracted, varimax rotation was used with Kaiser-Meier-Olkin (KMO) normalization (.93), indicating excellent factorability. KMO normalization measures whether or not a sample is adequate to determine matrix factorability. A KMO measure of at least .5 is acceptable and .8 and higher is considered good (Cerney and Kaiser 1977). Six components emerged with eigenvalues greater than 1 that explained 65% of the variance in the sample. A Cronbach's alpha reliability test was then performed in order to assess internal consistency of the six components. With the scoring of the CADI identical to Nolan and Grant's original version, six components were identified and labeled: difficulties in care relationship, restrictions on social life, family problems, financial consequences, physical care, and unsupportive services.

For the purpose of this study, the dependent variable, level of perceived caregiver burden, is operationalized as the composite score of all 30 individual CADI items, scored identically to those by Nolan and Grant (1992) and McKee and colleagues (2009). First, each of the thirty items was generated as a separate variable with a possible score range of 0-3. There were four available responses to each statement: 1) "This statement does not apply to me" with a coded value of 0, 2) "This statement applies to me and I find it not stressful" with a coded value of one, 3) "This statement applies to me and I find it stressful" with a coded value of 2, and 4) "This statement applies to me and I find it very stressful" with a coded value of 3. Each respondent in the sample was also assigned a total CADI index score, a continuous variable generated as a sum of all scores from the thirty individual CADI items. Possible index scores ranged from 0-90, with higher scores indicating greater perceived burden.

Independent Variable - Affiliate Stigma

Perceived affiliate stigma was measured using a six-item affiliate stigma scale adapted and refined from the three-item scale used by researchers examining stigma experienced by mothers of children with epilepsy in Zambia (Elafros et al. 2013). Their scale was actually an adaptation of an adaptation. Jacoby (1992) used Hyman's (1971) Short Stigma of Stroke format in developing a three-item stigma scale for patients with epilepsy. Elafros and her team further adapted this measure, not to measure the stigma from the PWE's point of view but to measure courtesy stigma experienced by the patients' mothers. Their 3-item scale was comprised of the following statement with three parts: "Because of my child's epilepsy: 1) some people are uncomfortable with me/my child, 2) some people would prefer to avoid me/my child, and 3) some people treat me/my child like an inferior person." For each statement, if the respondent agreed the response had a coded value of one (1). If the respondent did not agree the response had a coded value of zero (0). The possible scale scores ranged from 0-3. There was no differentiation between the stigmas the mothers perceived to be directed toward their children from the stigmas they perceived directed toward them personally. They also only studied mothers of pediatric epilepsy patients. Through e-mail correspondence, Ms. Elafros gave her approval to use or adapt the scale.

Epilepsy stigma measures in this study differentiated between the stigma familial caregivers perceived directed towards them from the stigma directed towards their family members, then combined the two into a total stigma score as a way to operationalize affiliate stigma. As Mak and Cheung (2008) noted, in affiliate stigma, the stigma directed toward the family member with the stigmatizing condition is perceived, acknowledged, and internalized by the unaffected family member resulting in a psychological response. To that end, the scale was refined by expanding the question into six parts and creating a six-item affiliate stigma scale. Wording was changed from “my child” to “my family member” in order to accommodate all ages of PWE and familial relationships other than parent/child. Three items measured the stigma the caregivers felt was directed toward their family member, each with a “yes” or “no” response choice: “Because of my family member’s epilepsy: 1) some people are uncomfortable with my family member, 2) some people would prefer to avoid my family member, and 3) some people treat my family member like an inferior person.” Another three items measured the stigma the caregivers felt was directed toward themselves: “Because of my family member’s epilepsy: 1) some people are uncomfortable with me, 2) some people would prefer to avoid me, and 3) some people treat me like an inferior person. Each individual item was scored 1 for “yes” and 0 for “no” responses. The family caregivers’ perceptions of stigma directed toward their family members with epilepsy had a possible range of 0-3, the possible range for stigma directed toward them was 0-3, and the total affiliate stigma scores had a possible range of 0-6. Higher scores were reflective of greater feelings of stigmatization. Three continuous variables were created in the dataset to reflect the three scale scores.

Independent Variable – Age of PWE

Ages of the respondents' family members with epilepsy were collected with the question, "How old is your family member today?" and recorded as continuous variables then re-coded into an additional dichotomous age-category variable. The decision was made to differentiate between children and adults using the age of 20 because special needs children in Alabama are allowed to attend public schools and receive services through this age. Upon turning 21 years old, they are no longer eligible to receive education services through the public school system (AAC 2013). Accordingly, if the patient was <21 years they were coded 0 as "pediatric", if 21 years or older, they were coded 1 as "adult" with pediatric as the reference category.

Independent Variables - Control

As control variables, additional demographic information was collected. Data regarding the gender of the PWE and participation in the CBD study were collected. Additionally, gender, age, marital status, race/ethnicity, education level and annual income of each caregiver were collected. Patients' gender data were ascertained with the question "What is your family member's gender?" with two response categories: male and female. According to the Institute of Medicine, which informs federal policy in data collection for the sciences, sex and gender data should be clarified (Institute of Medicine 2001). According to their guidelines, "sex" is a classification term based on reproductive organs and chromosomal differences while “gender” is a person’s self-representation or presentation. Both terms use a dichotomous classification of “male” and “female”. Recommendations also suggested data should be consistently gathered and reported. For this study the term “gender” was used in lieu of the term “sex” because with the self-administered nature of the questionnaire, there was no way to verify a respondent’s sex nor the sex of their family member. With this dichotomous variable, males were coded as 1 and females were coded as 0. “Female” was the reference category. In collecting caregivers’ genders, they were asked, “What is your gender?” with options “male” and “female” offered. Again, this gender variable was dichotomous, coded with males as 1 and females as 0 with “female” as the reference category.

Inclusion of the family member with intractable epilepsy in the CBD study was ascertained with the question, “Is your family member enrolled in the Alabama CBD Study? (Choose 1)”. Possible responses included “yes”, coded as 1, “no”, coded as 2, and “they were enrolled but are no longer in the study”, coded as 3. A dichotomous variable was generated to reflect those currently in the CBD study coded as 1 and those not in the study or having left the study coded as 0. Caregiver age was determined with an open ended question, “What is your age?” with a blank to enter years and was recorded as a continuous variable.

Race and ethnicity data were ascertained with the question “What category best describes your race/ethnicity? (Please choose up to 2).” Categories included: “white” coded as 1, “black or African American” coded as 2, “Hispanic or Latino” coded in a separate variable, “Asian” coded as 3, “American Indian or Alaska Native” coded as 4, and “Native Hawaiian or Other Pacific Islander” coded as 5. In an effort to be more inclusive, race and ethnicity were combined into one question with the opportunity to choose two items in order to provide an option for biracial participants and to provide Hispanic or Latino participants a chance to choose that category as their identified race. This decision was informed by several Hispanic and Latino participants in previous studies who expressed disappointment and irritation that they could not mark their ethnicity as their race. When these data were entered for analyses, an additional variable was created to record ethnicity with two categories: 1) Hispanic/Latino origin, coded as 1 and 2) non-Hispanic/Latino origin, coded as 0. Any respondent entering a single race as "Hispanic/Latino" were placed in the "white" race category and designated as "Hispanic/Latino origin" in accordance with the Food and Drug Administration's (FDA) recommendations for Collection of Race and Ethnicity in Clinical Trials (2005). A dichotomized race variable was generated with white having a value of 1 and all others (nonwhite) having a value of 0.

Data regarding caregivers' marital statuses, education levels, income levels, and number of residents in their homes were gathered as well. Marital status was measured with the following question "What is your marital status?" Response categories included: a) never married/in a committed partnership, coded as 1, b) married/in a committed partnership, coded as 2, c) divorced, coded as 3, d) widowed, coded as 4, and e) separated, coded as 5, with "married/in a committed partnership" as the reference category. Caregiver educational level was determined by asking "What is the highest level of education that you have completed? (Check one)." Response categories included: a) did not graduate high school, coded as 1, b) high school graduate or GED, coded as 2, c) vocational or trade school graduate, coded as 3, d) some college or associates degree (2-year degree), coded as 4, e) bachelor’s degree (4-year degree), coded as 5, and f) more than a 4-year degree, coded as 6. The reference category was “high school graduate/GED.” This ordinal variable was collapsed into two education variables. First, respondents who answered “did not graduate high school” and “high school graduate or GED” were combined into a category labeled “high school or less”, coded as 1. Respondents who graduated from vocational or trade school and those with some college or an associate’s degree were combined into a category labeled “some college”, coded as 2. Those who earned a bachelor’s degree or higher were combined into the final category labeled “bachelor’s degree or more”, coded as 3. The reference category was “high school or less.” A separate dichotomous education variable was created to reflect respondents with at least a bachelor’s degree, coded as 1 and less than a bachelor’s degree coded as 0 (reference group).

Annual household income level was assessed with the question: “What is your annual income level? (Check one).” This variable contained fifteen mutually exclusive categories offered in increasing increments starting with “less than $5,000”, coded as 1, “5,000 – 9,999”, coded as 2, “10,000 – 14,999”, coded as 3, “15,000 – 19,999”, coded as 4, “20,000 – 29,999”, coded as 5, “30,000 – 39,999”, coded as 6, “40,000 – 49,999”, coded as 7, “50,000 – 64,999”, coded as 8, “65,000 – 74,999”, coded as 9, “75,000 – 99,999”, coded as 10, “100,000 – 124,999”, coded as 11, “125,000 – 149,999”, coded as 12, “150,000 – 174,999”, coded as 13, “175,000 – 199,999”, coded as 14, and ending with “$200,000 or more”, coded as 15. This variable was operationalized so that the variable could be treated as ordinal by using these categories or continuous by taking the median dollar amount of each category and coding the response with that number then using a Pareto curve for the last category ($200,000 or more) (Ligon 1989). Finally, caregivers were asked the number of adults and children living in their homes with the open-ended questions “How many adults are living in your house?” and “How many children are living in your house?” These responses were recorded as continuous variables. A continuous variable was then generated to reflect the number of individuals in each home by adding the adults and children responses.

Two questions allowed for respondents to freely express themselves. “What services do you feel would be most helpful to you and your family?” was the first. Several lines were included to encourage responses. The second question was more of a statement, “Please use this space to comment on anything you’d like to add.” There were several lines for writing and the back of the survey was blank if the respondent needed more space.

Analytical Strategy

Univariate, CADI reliability, bivariate, and multivariate analyses were performed using the software package Stata 13.1 (StataCorp 2013). Data from the surveys were entered into a dataset created for this study named “CIE” (Caregiving in Epilepsy). During data cleaning, cases with missing data were dropped, as well as any case that answered “no” to the question “Are you a primary caregiver to a family member with intractable epilepsy? (Check one).” Responses from caregivers less than 18 years of age were dropped from the study as well, as they were not eligible per the IRB protocol.

Once additional variables were generated, univariate analyses were performed. Each variable’s distribution was examined for limitations, measures of central tendency and spread, and descriptive statistics were calculated to report demographic data. A

Cronbach's alpha score (α) was calculated for the total CADI score variable.   The resulting dataset was saved to use in bivariate and multivariate analyses.

Bivariate analyses included Pearson's correlation coefficients (from here on called correlations), independent sample *t*-tests, and one-way analysis of variance (ANOVA) tests. Correlations, with two-tailed significance tests, were used to assess the strength and direction of linear relationships between the scale control variables (age of the PWE and caregiver income), the total CADI scores, and scores the three stigma scales. Scatterplots were graphed and examined. ANOVAs and *t*-tests were used to assess the differences in means of stigma and CADI scores between the different groups in the categorical demographic variables, including the gender and categorical age variable of the PWE and the gender, education level, race, and marital status of the caregivers. Independent sample t-tests were conducted between the variable of CBD study enrollment status (participant/non-participant) and total CADI scores, total stigma, stigma perceived about the caregiver, and stigma perceived about the PWE. Cross-tabulations were completed to identify statistically significant associations between CBD study participation status and each of the 30 individual CADI items with chi-square tests of significance.

Finally, multivariate analyses were used to test the hypotheses. Four nested ordinary-least-squares (OLS) regression models were estimated with an *F*-test to determine which model had the best fit. Collinearity between independent variables was assessed with a variance inflation factor (VIF) score. The two hypotheses tested indicated direction in expected outcomes. While it was entirely appropriate to use one-tailed significance tests for the regression models in these analyses, two-tailed tests were conducted in order to mitigate type I errors. Significance was set at $\alpha = .05$.

In Model 1, the estimated model used the CADI total index score variable as the dependent variable with the total stigma score as the main explanatory variable. In Model 2, the regression model from above was again estimated, this time adding in the categorical age variable (pediatric/adult). Model 3 was then estimated controlling for the following demographic variables: caregiver age (continuous), number of people in the household (continuous), race of caregiver (dichotomized white/nonwhite), whether or not their family member was in the CBD study (dichotomized), the log of caregiver income level (continuous), and whether or not the caregiver had at least a bachelor’s degree (dichotomized). The log of annual income was used because the distribution of the income variable as it is with actual values was positively skewed. Taking the log normalized the distribution somewhat. Finally, in Model 4 the interaction term between the total stigma score and age category was added.

To summarize, this analytical strategy was designed to determine support or nonsupport for the three hypotheses stated in Chapter 3. The first hypothesis, which argued that there is a positive relationship between perceived epilepsy stigma and perceived burden among caregivers, would be considered supported if the coefficient for the stigma variable was positive and significant in Model 1. The second hypothesis, which stated that the relationship between perceived epilepsy stigma and perceived caregiver burden was mediated by the age of the family member with epilepsy, would be considered supported if the coefficient for age category was positive and statistically significant while the coefficient for total perceived stigma decreased or became not significant. For the third hypothesis, which stated that the relationship between perceived epilepsy stigma and perceived burden is moderated by the age of the family member with epilepsy, the hypothesis would be considered supported if the coefficient for the interaction term was positive and significant in Model 4.

Information from the open-ended questions in the survey was gathered for two primary reasons. First, data were collected to inform the grant proposal of perceived caregiver needs. Comments were also gathered to use as anecdotal evidence when describing the experience of caregiving in this population. A few verbal comments were recorded later in a notebook, in private, after the encounter in a non-identifying way in order to preserve confidentiality.

CHAPTER 5

RESULTS

In this chapter, results from univariate, bivariate, and multivariate analyses are presented. Univariate analyses include demographic results of caregivers and the family members for whom they provide care. Descriptions of CADI and stigma results are also included. Bivariate results from correlations, crosstabulations, and t-tests are also presented. The chapter ends with multivariate results from four nested OLS regression models.

Descriptive Analyses

Demographic Results

Demographic data were gathered for both the caregivers responding to the survey and the family members with epilepsy for whom they cared (Table 1). The average age of PWE was almost 23 years with 45% over the age of 20. Ages ranged from approximately 2 years old through 82 years. Males comprised 52% of the PWE in the sample and just over 60% were enrolled in the CBD study at the time of the survey.

Caregivers ranged in age from 22 through 83 with an average age of 48 years. Respondents were predominantly female (75%) and married (69%). The racial composition of the sample was primarily White (83%) and no Hispanics were

Table 1. Demographic Results (*N* = 136)

Variable	Mean/Freq	SD	Range
Family Member			
Age in years	22.63	16.56	2-82
% Adult	44.85		
% in CBD study	60.29		
% Male	52.21		
Caregiver			
Age in years	48.13	12.45	22-83
% Male	25.00		
Annual Income in dollars[a]	80,569.40	68,939.77	2,500-280,000
< 5,000	4.00		
5,000 – 9,999	8.00		
10,000 – 14,999	11.00		
15,000 – 19,999	8.00		
20,000 – 29,999	16.00		
30,000 – 39,999	16.00		
40,000 – 49,999	7.00		
50,000 – 64,999	12.00		
65,000 – 74,999	10.00		
75,000 – 99,999	12.00		
100,000 – 124,999	11.00		
125,000 – 149,999	7.00		
150,000 – 174,999	2.00		
175,000 – 199,999	5.00		
> 200,000 (Mean=280,000)	8.00		
Race/Ethnicity			
% White	83.10		
% Black	13.97		
% Am Indian/Alaska Nat	2.21		
% Asian	.74		
% Hispanic	0.00		
Marital Status			
% Married	69.12		
% Divorced	15.44		
% Never married	7.35		
% Widowed	4.41		
% Separated	3.68		
Education Level			
% < High School	7.35		
% HS diploma	22.06		
% Vocational diploma	3.68		
% Some college	34.56		
% Bachelor's Degree	15.44		
% > Bachelor's	16.91		

[a] Annual income used as logged variable in analyses

represented. Education levels varied greatly with almost 35% reporting attending some college or earning a two-year degree. Respondents with a high school diploma (22%) were second in frequency and 17% had earned more than a bachelor's degree. Income levels also varied greatly. When converted to a continuous variable with a value of $280,000 assigned to the top income category, the average annual income was $80,000 with 23% reporting an annual household income between $20,000 and $40,000. Another 23% had less than $20,000 coming in each year.

CADI Results

Individual items as well as the total score of the CADI index were analyzed for measures of central tendency (Table 2). With a possible total score range of 0-90, the total CADI scores had a mean of 33.63, with a Cronbach's alpha score of .95. The distribution was positively skewed with a median score of 29. For individual items, "It is physically tiring" had the highest mean score (1.75 out of a possible 3) and "I no longer have a meaningful relationship with the person I care for" scored the lowest (.24).

Item response frequencies were analyzed as well (Table 3). The item least associated with this sample was "I no longer have a meaningful relationship with the person I care for" with 115 respondents reporting it did not apply to them. Of the 21 respondents who felt this applied to them, over half did not feel stressed by it. Of the 26 who reported no satisfaction from caring, 15 did not feel troubled by that feeling.

Table 2. Carer's Assessment of Difficulty Index (CADI) (*N* = 136)

Index	Mean (SD)	Range	α
Total CADI Score	33.63 (20.36)	0-90	.95
<u>Individual Items</u>			
I can feel helpless/not in control of the situation.	1.64 (.98)	0-3	
My emotional well-being suffers.	1.41 (1.00)	0-3	
It can put a strain on family relationships.	1.60 (1.04)	0-3	
I can't relax because of worry about caring.	1.57 (1.03)	0-3	
I don't have enough private time for myself.	1.45 (1.09)	0-3	
I can't devote enough time to other family members.	1.37 (1.04)	0-3	
I feel angry about the situation.	.85 (1.02)	0-3	
I feel guilty about the situation.	.94 (1.11)	0-3	
It is physically tiring.	1.75 (1.03)	0-3	
The person I care for is immobile/has problems getting about.	1.03 (1.19)	0-3	
The person I care for doesn't always help as much as they could.	1.03 (1.05)	0-3	
My physical health has suffered.	1.0 (1.12)	0-3	
The person I care for needs a lot of help with personal care.	1.40 (1.10)	0-3	
My sleep is affected.	1.46 (1.10)	0-3	
The person I care for is incontinent.	.90 (1.10)	0-3	
It restricts my social life/outside interests.	1.55 (1.09)	0-3	
I can't have a break or take a holiday.	1.21 (1.09)	0-3	
I can't see friends as often as I'd like.	1.18 (1.05)	0-3	
Professional workers don't seem to appreciate the problems caregivers face.	1.10 (1.07)	0-3	
I don't get enough help from the health and social services.	1.20 (1.18)	0-3	
It causes financial difficulties.	1.51 (1.15)	0-3	
My standard of living has fallen.	.99 (1.17)	0-3	
Relatives don't keep in touch as often as I'd like.	.93 (1.12)	0-3	
Some family members don't help as much as they could.	1.28 (1.20)	0-3	
The person I care for sometimes manipulates me.	.60 (.97)	0-3	
The person I care for can demand too much of me.	.81 (1.01)	0-3	
I no longer have a meaningful relationship with the person I care for.	.24 (.64)	0-3	
The person I care for doesn't always appreciate what I do.	.60 (.85)	0-3	
The behavior of the person I care for is a problem.	.75 (1.04)	0-3	
There is no satisfaction to be gained from caring.	.30 (.70)	0-3	

Table 3. Frequencies of Individual CADI Items (*N* = 136)

Item	*NA	*NS	*S	*VS
I can feel helpless/not in control of the situation.	21	35	52	28
My emotional well-being suffers.	37	21	63	15
It can put a strain on family relationships.	28	28	51	29
I can't relax because of worry about caring.	26	36	45	29
I don't have enough private time for myself.	36	31	41	28
I can't devote enough time to other family members.	37	33	45	21
I feel angry about the situation.	69	33	20	14
I feel guilty about the situation.	66	34	14	22
It is physically tiring.	19	36	41	40
The person I care for is immobile/has problems getting about.	67	25	17	27
The person I care for doesn't always help as much as they could.	58	31	32	15
My physical health has suffered.	66	23	28	19
The person I care for needs a lot of help with personal care.	36	39	31	30
My sleep is affected.	34	37	34	31
The person I care for is incontinent.	71	26	21	18
It restricts my social life/outside interests.	30	35	37	34
I can't have a break or take a holiday.	48	32	35	21
I can't see friends as often as I'd like.	48	31	41	16
Professional workers don't seem to appreciate the problems caregivers face.	53	34	31	18
I don't get enough help from the health and social services.	57	22	30	27
It causes financial difficulties.	37	29	34	36
My standard of living has fallen.	69	25	17	25
Relatives don't keep in touch as often as I'd like.	70	26	20	20
Some family members don't help as much as they could.	52	25	28	31
The person I care for sometimes manipulates me.	93	14	20	9
The person I care for can demand too much of me.	74	25	26	11
I no longer have a meaningful relationship with the person I care for.	115	12	6	3
The person I care for doesn't always appreciate what I do.	81	34	15	6
The behavior of the person I care for is a problem.	82	19	22	13
There is no satisfaction to be gained from caring.	110	15	7	4

*NA=Does not apply to me, NS=Not Stressful, S=Stressful, VS=Very Stressful

The acknowledgment that caregiving is physically tiring was the item most reported as applicable to respondents. Only 16% said it did not apply to them while 69% of those who said it applied to them found it stressful or very stressful. Ambulatory issues affected 51% of the PWE in the study and 64% of their caregivers found this stressful. Incontinence was a problem for 48% with 60% of their caregivers finding it stressful or very stressful.

Of those reporting the item relevant to them, emotional upset was a frequent occurrence with more than 70% feeling helpless and emotionally strained and 50% admitting feeling guilty or angry about the situation. Financial costs related to caregiving were assessed with “It causes financial difficulties” and “My standard of living has fallen”. Around 72% of respondents felt stressed or very stressed with this indicator of burden while 63% were troubled that their standard of living had fallen. Issues with extended family were problematic. Sixty-one percent felt upset that relatives no longer kept in touch and 53% were stressed that family members did not help out more.

Perceived Stigma Results

The six items in the affiliate stigma scale were analyzed individually and collectively (Table 4). The overall scale score for total stigma had a possible range of 0-6 and had a mean of 2.79. With a possible range of 0-3, the scale measuring perceived stigma of the family member with epilepsy had a mean of 1.71 with a negatively skewed distribution. Of the three items that comprise this measure, just over 74% felt that people are uncomfortable being around their family member, 50% believed people avoid their family member, and 47% reported their family member being treated as an inferior person. The scale measuring perceived stigma of the caregiver had a mean of 1.08 with apositively skewed distribution. When looking at each of the three individual items in this measure, over 50% reported that people felt uncomfortable around them because of their family member with epilepsy with 35% feeling that people avoided them. Around 22% felt they were treated as an inferior person.

Table 4. Perceived Epilepsy Stigma Scales ($N = 136$)

Index	Mean	Range
Perceived Stigma of Family Member	1.71	0-3
Perceived Stigma of Caregiver	1.08	0-3
Total Stigma Score	2.79	0-6
Individual Items		% Yes
People are uncomfortable around me		50.74
People avoid me		35.29
People treat me like an inferior person		22.06
People are uncomfortable around my family member		74.26
People avoid my family member		50.00
People treat my family member like an inferior person		47.06

Bivariate Associations

Correlations were performed between the total CADI score, perceived stigma of the caregiver, perceived stigma of the PWE, the age of the PWE, caregiver age, and annual household income (Table 5). There was a moderate positive relationship between the CADI scores and both the perceived stigma of the caregiver (.49) and the perceived stigma of the PWE (.47) with both significant at $p<.001$. Between the two stigma scales, there was a strong positive relationship (.70) that was significant at $p<.001$. Perceived stigma of the caregiver also had weak negative relationships with caregiver age (-0.18) and PWE age (-0.17), both significant at $p < .05$. Income was not significantly associated with any of the included variables.

Table 5. Correlations of Interest (*N* = 136)

	A	B	C	D	E	F
Total CADI	1					
Stigma Caregiver	0.49***	1				
Stigma PWE	0.47***	0.70***	1			
PWE Age	0.01	-0.17*	-0.10	1		
Caregiver Age	-0.04	-0.18*	-0.07	0.61***	1	
Income	-0.15	-0.12	-0.15	-0.14	-0.04	1

*p < .05, ***p < .001

Independent sample *t*-tests were conducted between the continuous variables of CADI total scores, total stigma scores, PWE stigma, and caregiver stigma and the two-category variables gender of PWE, race of caregiver (dichotomized white/non-white), and age of PWE (dichotomized pediatric/adult with adults being age 21+ years). There were no statistically significant bivariate associations between them except for the categorical age variable (Table 6). There was a statistically significant difference (p < .05) in both the perceived stigma of the caregiver and the mean total stigma scores between caregivers of children under 21 years old and those who care for adult family members.

Table 6. Independent Sample *t*-tests for Age Categories (*N* = 136)

	Pediatric <21 years		Adult 21+ years		
	Mean	SE	Mean	SE	*t*-statistic
CADI Total	32.51	2.03	35.02	3.00	-0.71
Stigma Total	3.13	.26	2.38	.27	2.03*
Stigma PWE	1.83	.13	1.57	.16	1.21
Stigma Caregiver	1.31	.14	.80	.13	2.57*

*p < .05 *df*=134

When looking at whether or not the family member with epilepsy was enrolled in the CBD study, independent sample *t*-tests were conducted comparing total CADI scores, total perceived stigma, and the perceived stigma of the caregiver and PWE by enrollment status (Table 7). There were statistically significant differences between those enrolled in the CBD study and those not enrolled for all four variables. Caregivers of family members in the CBD study reported more total burden, more total stigma, and higher levels of both stigma towards them and their family member with epilepsy.

Table 7. Independent Sample *t*-tests for CBD Enrollment (*N* = 136)

	Participant		Nonparticipant		
	Mean	SE	Mean	SE	*t*-statistic
CADI Total	38.09	1.96	26.87	3.04	-3.25**
Stigma Total	3.18	0.24	2.20	0.28	-2.61**
Stigma PWE	1.88	0.12	1.46	0.17	-1.98*
Stigma Caregiver	1.30	0.14	.74	0.13	-2.84**

*$p \leq .05$, **$p \leq .01$ $df = 134$

ANOVAs were performed between marital status and the total CADI scores, total stigma, perceived stigma of the PWE, and perceived stigma of the caregiver. There was no evidence of significant differences between the marital categories in caregiver burden or perceived stigma. The same was done for caregiver race and caregiver education. Again, there was no evidence of differences between categories for either caregiver burden or stigma in either. When an ANOVA was performed between CADI scores and the categorical income variable, there was evidence of a significant difference between the income categories ($F = 2.02$ *df* (14), $p < .05$).

Finally, cross-tabulations were conducted between the CBD study status and each of the 30 individual CADI items. Half of the individual items were significantly associated with being in the CBD study (Table 8). Caregivers with family members in the CBD study were more likely to find the CADI item applicable to them. Those who found these 15 aspects of caregiving applicable were also more likely to feel stressed or very stressed by each item.

Table 8. Significant Cross-tabulations between CBD Study Status and Individual CADI items (*N* = 136)

Item	χ^2
I can feel helpless/not in control of the situation.	11.60**
My emotional well-being suffers.	10.38*
I can’t relax because of worry about caring.	16.48**
I don’t have enough private time for myself.	13.94**
I can’t devote enough time to other family members.	13.81**
It is physically tiring.	14.61**
The person I care for is immobile/has problems getting about.	16.27**
My physical health has suffered.	11.90**
The person I care for needs a lot of help with personal care.	18.24***
My sleep is affected.	12.64**
It restricts my social life/outside interests.	12.68**
I can’t have a break or take a holiday.	22.47***
I can’t see friends as often as I’d like.	10.98*
Professional workers don’t seem to appreciate the problems caregivers face.	15.23**
Some family members don’t help as much as they could.	9.70*

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

Multivariate Analyses

Four nested OLS regression models were estimated to predict total CADI scores (Table 9). Standardized betas are presented in Table 10. In determining which social variables to include in the models, several experimental nested regression models were estimated with collapsed and expanded demographic variables to identify the most efficient ways to code the variables for the best fit. Ultimately the following variables were used: the dichotomized PWE age variable (pediatric<21, adults 21+ years), caregiver age (continuous), number of people in the household (continuous), dichotomous race (White/nonwhite), whether or not the patient was in the CBD study, log of income (continuous), and if the caregiver had earned at least a bachelor's degree (dichotomous). A VIF test was performed to check the variables for collinearity. The mean VIF was 1.78 and no individual variable had a score more than 3.07 which precluded the need to center the continuous variables.

Table 9. Nested OLS Regression Models Predicting Total CADI Scores

	Model 1		Model 2		Model 3		Model 4	
	β	SE	β	SE	β	SE	β	SE
Total Stigma	4.85***	.69	5.10***	.69	4.66***	.70	3.50***	.90
Adult PWE			6.37*	3.01	7.77*	3.65	0.56	5.06
Caregiver Age					-0.07	.15	-0.09	.15
# in Household					0.74	1.04	0.70	1.03
White					-4.37	4.12	-4.32	4.06
In CBD Study					7.71*	3.09	8.13**	3.06
Income (log)					-3.07	1.63	-3.16	1.61
≥Bachelor's					4.58	3.48	4.92	3.44
Stigma*Adult PWE							2.74*	1.35
Constant	20.08	2.43	16.53	2.93	48.81	18.78	53.82	18.72
F	49.93***		27.84***		8.95***		8.61***	
(df)	(1, 134)		(2, 133)		(8, 127)		(9, 126)	
R^2	.27		.30		.36		.38	
Nested F	-		4.46*		2.17		4.12*	

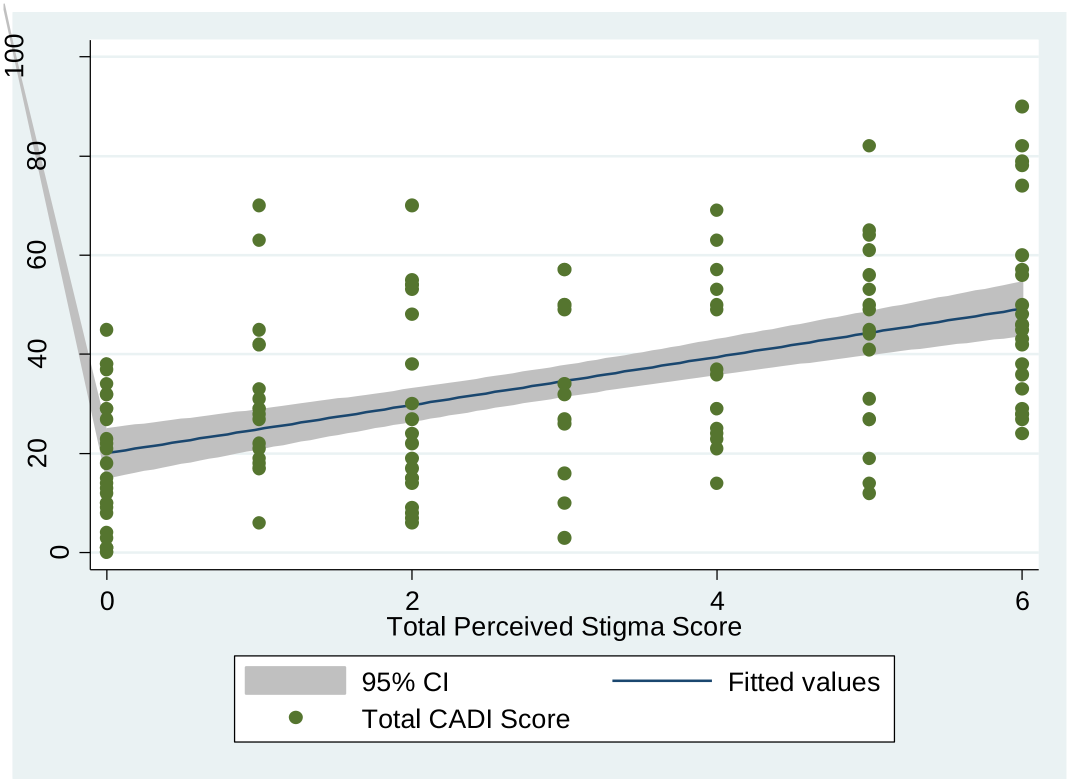
**p* < .05; ***p* < .01., ****p* < .001, (*N* = 136)

Table 10. Standardized Betas for Regression Models

	Model 1	Model 2	Model 3	Model 4
Total Stigma	.521	.548	.501	.376
Adult PWE		.156	.190	.014
Caregiver Age			-.044	-.053
# in Household			.057	.053
White			-.081	-.080
In CBD Study			.186	.197
Income (log)			-.157	-.162
≥Bachelor's			.106	.113
Stigma*Adult PWE				.246

In Model 1, the total burden score was regressed on total stigma score producing an *F* statistic of 49.93 (*df* 1, 134) that was statistically significant at *p* < .001. For every one point increase in stigma score, the total burden score increased by 4.85 (*p* < .001). When considered as the only predictor, affiliate stigma explained 24% of the variation in caregiver burden scores. Figure 4 provides a visual representation of the regression line.

Figure 4. Regression Line for DV-CADI Scores, IV-Stigma Scores (*N* = 136)



Model 2 was estimated with an additional age categorical variable, where adults were considered to be 21 years of age or older. This model as a whole yielded an *F* statistic of 27.84 (*df* 2, 133) which was significant at *p* < .001. The effect of perceived stigma on caregiver burden was not ameliorated by whether the PWE was an adult or a child when only looking at these two independent variables. Instead there was a slight suppression effect. When age category was added to the model to test for mediation, the coefficient for stigma increased rather than decreased, with both of the predictors statistically significant. For each one point increase in stigma score, burden level increased by 5.10 ($p < .001$), net of controls. Compared to caring for children, providing care for adult family members with epilepsy increased burden scores by 6.37 ($p < .05$) holding total perceived stigma constant. Adding age category as a predictor helped to explain the variation in burden perception significantly. Thirty percent of the variation in burden was explained by perceived stigma and age category of the PWE and the change in R^2 from the first regression model was significant (nested $F = 4.46$, $p < .05$).

When caregiver age, number of people in the household, dichotomized race, whether the PWE was in the CBD study, income, and whether or not the caregiver had at least a bachelor’s degree were added to the third model, 36% of the variation in burden scores was explained, with three statistically significant independent variables. The log of the continuous income variable was used. By taking the log of income, the distribution was more normal and made for a better model fit. In this case, for every one point increase in perceived stigma there was a 4.66 point increase in burden, net of controls, and this was statistically significant at $p < .001$. Compared to caregivers of children, caregivers of adults scored 7.77 points higher on the burden scale net of controls ($p < .05$). Finally, compared to those caregiving for patients not in the CBD study, those with family members in the study scored 7.71 points higher on the burden index, net of controls, and this was also statistically significant at $p < .05$. The model as a whole was statistically significant ($F = 8.95$, *df* 8, 127).

Age of the family member with epilepsy had a moderating effect on the relationship between total perceived stigma and burden levels. When stigma levels were interacted with age category (reference category is pediatric) in the final regression model, the coefficient of the interaction term ($\gamma = 2.74$) was statistically significant ($p < .05$). For every one unit increase in stigma, burden level increases 6.24 for caregivers of adults and by 3.50 for those caring for children. There was one other significant predictor in this model. Compared to those not in the CBD study, caregivers of patients in the CBD study had an increase of 8.13 points on the burden scale, significant at $p < .01$, net of controls. Not only was the final model statistically significant at $p < .001$ ($F = 8.21$), the model as a whole explained 38% of the variation in burden scores. The nested *F*-test ($F = 4.12$ $p < .05$) indicated that the R^2 change was also significant and this model had the best fit of the four.

In summary, this chapter contained the results from quantitative analyses for this study. As indicated in the analytical strategy in Chapter 4, results from the two qualitative-style questions about the needs of caregivers and additional comments were not included here but rather in Chapter 6 as part of the discussion.

The first hypothesis, that there is a positive relationship between affiliate stigma and perceived caregiver burden, is supported with 27% of the variation in burden levels explained by levels of perceived stigma. The second hypothesis, that age mediates the relationship between affiliate stigma and levels of perceived burden, is not supported. However, Model 2 indicates that 30% of the variation in burden is explained by affiliate stigma perceived by the caregiver and the age of the PWE. Rather than ameliorating, age had a slight suppression effect and the overall model was still statistically significant, most likely due to the higher stigma found among caregivers of children. The last hypothesis, that age of the PWE moderates the relationship between affiliate stigma and caregiver burden, is supported. While caregivers of children reported higher levels of stigma on average, stigma had a stronger impact on CADI scores for adult caretakers. When the interaction term between stigma and age category was added in Model 4, the coefficient for the interaction term was positive and statistically significant; the association between stigma and burden is stronger among those who care for adult PWE compared to caregivers of children. Also, this model had the best fit of the four with 38% of the variation in perceived levels of burden being explained.

CHAPTER 6

DISCUSSION AND CONCLUSION

This study provides an interesting glimpse into an aspect of caregiver burden that is rarely examined: that of how affiliate stigma might impact levels of burden. Considering that most studies of caregiver burden in epilepsy measure that burden with QOL checklists, which might or might not include a stigma component, this study is important in differentiating stigma from caregiver burden. The focus on affiliate stigma is also unique, especially as it is used as a predictor of caregiver burden. As well, the majority of other studies focus on either pediatric or adult populations. This study, on the other hand, examines both groups with an identical instrument in order to tease out variations in the intersection of stigma and burden perceived by familial caregivers.

Unsolicited feedback from caregivers taking the survey was encouraging in that several indicated no one had ever asked them how they felt about caregiving or about epilepsy stigma concerns. One woman who did not complete the survey stopped after the stigma scale items. Her child was younger than two years old and she thought the survey was “brutal.” This mother’s responses were not included in analyses because the CADI section was incomplete but her comment was written on the back of her survey with notes. Her child was enrolled in the CBD study and was physically disabled with developmental challenges. It was possible the survey reminded her of the stigma they might encounter as he grows up as well as the caregiving issues she may face along the way.

Others, though, took a more favorable view and thought the survey was helpful. One parent of a young son explained that, as a physician, she did not experience any stigma or caregiving burden because she was educated about epilepsy, but understood that her patients most likely would and was complimentary of the study. Several caregiving mothers admitted that no one had ever asked them about how they felt about caregiving or what they needed and were appreciative of the opportunity to comment.

Affiliate Stigma and Caregivers in Epilepsy

The decision to examine stigma was right on target based on the caregivers' responses in this study. In looking at the results from each of the individual stigma items, caregivers seemed to be acutely aware that they and their family members bear a stigma. With almost 75% reporting that people are uncomfortable around their family member and 51% saying others are uncomfortable around them, this level of discomfort could lead caregivers to use avoidance and social isolation as ways of coping with the stress of anticipated stigma. For example, several respondents commented that they purposely do not expose their family members to the outside world in order to protect them from rude looks and comments. One mother said of her 22-year old daughter, "She is always so happy. She doesn't know how mean and awful people can get and I'm not about to let her find out." Another mother said, "We just stay home; don't go anywhere. We used to go to church but he got to where he would scream and yell and people would act like they wanted us to leave, like it was my fault or something."

These instances of isolating or hiding in an attempt to manage the stigma they expected to receive are similar to impression management techniques in other health conditions such as morbid obesity (Hansen and Dye 2016), HIV (Siegel, Lune, and Meyer 1998), and leprosy (Barrett 2005). In these health conditions though, the affected person decides to hide from public view, while in intractable epilepsy the person making that decision to isolate is often a well-meaning family member. There is evidence of stigma coaching leading to the concealment of an epilepsy diagnosis (Schneider and Conrad 1980) but this can be difficult or nearly impossible with intractable epilepsy as seizures are more frequent and unpredictable. More than one family caregiver echoed one father who said, “Usually it’s just easier to stay in when he’s having a bad day. No one will stare at us at home.”

When considering the perception of stigma independently from how it relates to burden, more than half the caregivers surveyed reported that people avoid their family member with epilepsy and another 35% believe people avoid them as well, which does not bode well for families creating and maintaining social connections. With this study’s finding of a small but statistically significant negative correlation between the age of the PWE and the amount of perceived stigma toward the caregiver, it appears that as family members with epilepsy get older, the level of stigma perceived by their caregivers may decrease. This finding is in line with other studies’ results that the perception of epilepsy stigma by caregivers decreases over time (Leaffer et al. 2011; Rood et al. 2014). This may be due, in part, to some caregivers’ reduced exposure to other people in social situations due to social isolation. It may also be that the caregiver becomes used to societal reactions or perhaps they regard strange looks and comments as general interest on the part of the public actor rather than prejudice. As one mother of a 30-year old daughter said, “When she was little, everything people said and did bothered me. I stared people down, called them out, you name it. Now I just get out, do what I have to do at the store, and ignore everybody."

When considering the psychosocial aspect of affiliate stigma, there is evidence supporting the notion that the stigma perceived by familial caregivers in intractable epilepsy is affiliate stigma rather than courtesy stigma. The increased levels of caregiver burden reported by those who perceive higher stigma are indicative of the levels of stress this population is under. One mother of an adult son mentioned that when her son was younger the physical effects of his epilepsy were easier to deal with, as were the comments she would hear. As he got older and his seizures became more frequent, he lost interest in school and over time became more helpless and required more care. She felt the "looks" she would get from people in supermarkets and at church were more difficult to brush off as he became more visibly disabled.

Familial Caregiver Burden in Epilepsy

The CADI is an excellent instrument for measuring the different domains of caregiving. Results from this study's CADI scores illustrate the high internal consistency of the CADI instrument as a whole; the Cronbach's alpha of .95 was slightly higher than the original index designer's alpha of .92 (Nolan, Grant, and Keady 1998). As burden is no longer simply divided between subjective and objective aspects, the 30 items in the index cover an exhaustive list of activities, feelings, and situations that add to the stress in caring for someone with intractable epilepsy. Most burden studies in epilepsy use either a qualitative methodology with interviews or focus groups, or a checklist of health related

QOL issues, and the fact that stigma is not addressed in the CADI is a distinct advantage. Using this instrument makes it simpler to differentiate stigma from caregiving.

The frequency table of the individual item responses indicates that there is significant variation in burden for this population of caregivers. Considering the vast majority of the caregivers in this study are parents, and all are family members, it is not surprising that very few reported feeling like they do not have a meaningful relationship with their family member or that they feel no satisfaction in providing care.

The acknowledgment that caregiving is physically tiring was the item most reported as applicable to respondents. This is understandable because intractable epilepsy is frequently associated with intellectual and physical disabilities rendering some patients unable to independently perform activities of daily life such as walking, bathing, feeding oneself, and using the toilet (Kwan and Brodie 2000). As PWE grow out of childhood and into adulthood, they become larger and heavier, making it more difficult for caregivers to perform these activities. As well, sleep is affected for many caregivers in this study, similar to results from other studies. Reduced hours of sleep and fitful sleep due to worry about seizures are common occurrences (Gibson 2014; Smith et al. 2014). One mother in this study reported that when her child is having a difficult day with seizures, she sleeps in a sleeping bag on the floor next to her daughter's bed in case she needs her.

The CADI, in allowing respondents to choose the option "does not apply to me," offers a chance to examine the levels of stress associated with each item relative to other caregivers experiencing the stressor. There were a few respondents, however, who marked "does not apply" on every item in the index indicating no burden at all. One mother remarked, "[*Son's name*] is a gift from God. I was chosen to take care of him and it wouldn’t do for me to act like that was a problem.” Another parent remarked, “It would be nice to not worry about buying the new wheelchair but God will provide.” While religiosity was not measured in this study, there was a strong thread of religious faith running through the spoken and written comments made by those surveyed.

Of those reporting the item relevant to them, emotional upset was a frequent occurrence with more than 70% feeling helpless and emotionally strained and 50% admitting they feel guilty or angry about the situation. Financial costs related to caregiving were assessed with the CADI items “It causes financial difficulties” and “My standard of living has fallen.” Around 72% of respondents in this study felt stressed or very stressed with this indicator of burden while 63% were troubled that their standard of living had fallen.

Issues with extended family can be problematic for caregivers, especially when the PWE displays troubling behaviors (Wagner et al. 2009), and this study’s participants were no different. A cross-tabulation of the two CADI items “The behavior of the person I care for is a problem” and “It can put a strain on family relationships” yielded a statistically significant association (Table 11). Other studies have revealed problematic behaviors including temper tantrums (Thompson et al 2014), rage (Wagner et al. 2009), hyperactivity and self-injury (Nolan, Camfield, and Camfield 2006), and excessive masturbation (Ozmen et al. 2004).

This last behavior was particularly challenging for one of the parents in this study. When her son became an adolescent, he went through an extended period of time where he would self-stimulate at various times throughout the day. The school system in their

Table 11. Cross-tabulation of Problem Behaviors and Family Strain ~% (*N* = 136)

	The behavior of the person I care for is a problem			
It can put a strain on family relationships	NA	NS	S	VS
NA-Does not Apply	18%	1%	0%	1%
NS- Applies/Not Stressful	13%	5%	2%	0%
S-Applies/Stressful	20%	5%	10%	3%
VS-Applies/Very Stressful	9%	3%	4%	5%
$\chi^2 = 28.36$, $p < .01$				

rural southern county decided they were unable to accommodate his needs in their school and after the teachers pressured her, she decided to home-school him. While this decision resolved the school problem, it served to further isolate her. The time her child was at school had provided a much-needed respite for her. She and her sister would meet for lunch several times a week and the mother reported she felt “almost normal” at these times. Once her son was home all day every day, she tried to continue having lunch with her sister at her home but was not comfortable moving her son into another room while they ate. When her son would start masturbating, she would try to gently make him stop, but he would become combative, and she usually ended up covering his lap with a blanket. Her sister was embarrassed and unwilling to stay and insisted that her sister move her son to a nursing home. She refused to consider this and eventually her sister stopped visiting entirely. This is but one example of the myriad of unsolicited stories related by respondents while taking the survey reflective of the behavioral challenges faced by caregivers.

Intersection of Affiliate Stigma and Caregiver Burden

One of the primary assumptions of this study is that there is a positive relationship between affiliate stigma and perceived burden among caregivers in intractable epilepsy. Results from this study provide a strong argument that this may be the case. Because previous studies did not typically differentiate between stigma and burden, it has been unclear how much of an effect perceived stigma might have on burden. This is the first study to show a very strong and statistically significant association. Other studies also did not include caregivers of both children and adults. Unfortunately, this aspect is overlooked in most studies of caregiving in epilepsy. Considering the extensive physical and intellectual disabilities many persons with intractable epilepsy have, the needs of these patients and their caregivers change as they grow older and having data to compare across groups is important.

Evidence also supports the second assumption of the study which stated that the relationship between perceived epilepsy stigma and perceived burden varies between caregivers of adults and caregivers of children. While patient’s age (pediatric vs. adult) did not mediate the relationship between perceived stigma and a caregiver’s sense of burden, it did act as a moderator, strengthening the relationship, i.e., the association between stigma and CADI burden was stronger among caregivers of adult PWE compared to pediatric, and that was after factoring in the age, race, family size, household income, and education level of the caregivers. It is surprising that the number of people in a household was not significantly associated with caregiver burden scores. Intuitively it may seem that if there are more people in a home to take care of, the burden load would be greater for a primary caregiver or perhaps smaller families might not have others to share in their tasks and experience greater burden, but this did not seem to be the case.

It is unclear why caregiver age, race, education level, and income were not associated with total burden levels. Compared with non-white families, white caregivers reported lower burden scores, though the association was not significant. Further examination by cross-tabulating each of the thirty individual burden items with race yielded only one significant result (Table 12). Non-whites who found no satisfaction to be gained from caring were more likely to rate this aspect of caregiving as “very stressful” than whites. There was no statistical association between education level and total burden which may have been due to the relatively high level of education in the sample. While having a bachelor’s degree or higher had no significant effect on total burden, there was one individual burden item with a significant association (Table 13). Caregivers who had not earned a bachelor’s degree were more likely to rate their family member’s incontinence as “not stressful.”

While income was not a significant predictor of total burden levels, it was significantly associated with six different individual CADI items. Not surprisingly, the most significant of these associations were financial difficulties and lower standards of living. Poorer physical health, resentment toward family members for not helping more, irritation that social service workers do not understand the stress they are under and lack of meaningful relationship with their family member were also significantly related to income levels. There is evidence of varying levels of stigma associated with using social service programs. For example, there is more stigma associated with those using state programs such as Aid to Families with Dependent Children (AFDC), Temporary Aid to

Needy Families (TANF), and Medicaid than for those using federal guaranteed assistance programs such as Social Security Disability (SSD), and this stigma is negatively associated with income level, i.e., those with lower incomes experience higher levels of stigma (Williamson 1974; Jarrett 1996; Stuber and Kronebusch 2004; Allen et al. 2014). Future studies might consider the possibility of a double stigma perceived by lower-income caregivers in epilepsy and tease out the stigma they might perceive for having a family member with epilepsy from the stigma they might perceive due to having a low income and needing social services.

Table 12. Cross-tabulation of Race and "No Satisfaction from Caring" ($N = 136$)

	Race	
No satisfaction to be gained from caring	White	Non-White
Is not stressful	65%	33%
Is stressful	30%	17%
Is very stressful	5%	67%
Total	100%	100%
$\chi^2 = 9.94$, $p < .05$		

Table 13. Cross-tabulation of Caregiver Education and PWE Incontinence ($N = 136$)

	Education Level of Caregiver	
"Person I care for is incontinent"	< Bachelor's	Bachelor's or higher
Is not stressful	51%	18%
Is stressful	21%	55%
Is very stressful	28%	27%
Total	100%	100%
$\chi^2 = 9.94$, $p < .05$		

One interesting significant finding was that when a caregiver's family member was in the CBD study, they experienced more total burden. One possible explanation for this might be the high frequency of visits the PWE make to the research clinic for the study. Until patients are stable on a dosage of CBD, they are required to attend clinic every two weeks. For patients who are combative, non-ambulatory, or require significant assistance with personal care, this can be challenging for caregivers, especially those who drive several hours. Because many of the caregivers were recruited from the CBD study it is possible the caregivers were experiencing high levels of stress while completing the survey. There is also a possibility that seizure frequencies and severity may vary between those in the CBD study and those not participating. Some caregivers adamantly against using marijuana for medical purposes might have not considered participating in the study. These variations remain unclear and might be examined in future studies.

Limitations

There were a few limitations in this study concerned the study design. With the use of a convenience sample and the cross-sectional nature random sampling was not possible. A longitudinal study would have been more robust as it would examine variations in perceived levels of stigma and caregiver burden over time but this was not possible with the time constraints. Data concerning the severity and types of seizures patients experienced were not captured. This information would have been helpful in examining the differences between the group in the CBD study and those not enrolled. It is possible those in the study suffered more debilitating forms of epilepsy leading to higher levels of burden. It is also possible the group not enrolled in the CBD study were ineligible due to co-existing health or genetic issues. Future studies should include gathering seizure types and frequencies. Also, the survey was self-administered which may introduce bias.

The reliance on physicians to allow access to clinics in order to approach caregivers was helpful in that the survey was directed toward caregivers of those with intractable epilepsy rather than general epilepsy patients. This aspect also limited the eligible patient pool because very few physicians allowed access to their clinics. There was also little variation in race with respondents being mostly white with no Hispanic or Latino participants. Using data from an original survey is also a limitation in that the survey has not been validated from repeated use in other studies. There simply was not a dataset available for public use that measured both perceived stigma and levels of caregiver burden.

Implications for Research and Practice

This study is an important step in the research into stigma and caregiver burden in intractable epilepsy. Among the first studies to provide evidence of affiliate stigma among caregivers in intractable epilepsy, it adds to the academic literature from which to expand future research. Repeating this study will provide opportunities for validation of the survey instrument. Other researchers might utilize a mixed-methods approach, using the instrument as a quantitative measurement of affiliate stigma and caregiver burden and incorporate a few in-depth interviews or focus groups to flesh out the perceptions of stigma and to discuss the specific aspects of burden that are troubling.

Particular emphasis in future studies might examine the psychological responses in affiliate stigma exhibited by caregivers, such as isolating behaviors and other coping mechanisms. Before this study, affiliate stigma in intractable epilepsy had not been examined. In fact, it has not been examined in many health-related conditions. Viewing epilepsy through this lens can provide motivation to study affiliate stigma in other disorders.

Results from this study also add to the literature on caregiving in chronic illness and the stress process.

Neurologists who treat PWE might use these results to assist in identifying concerns of caregivers. With limited time spent during office visits, the needs of caregivers might be overlooked. Adequate space in waiting rooms and examination rooms for wheelchairs and caregivers is a need several caregivers expressed. Other items related to medical office environment were dim lighting, quiet areas to wait with few environmental triggers, and family style restrooms that accommodate large wheelchairs. Access to social services that are available in their area would be extremely helpful. Many caregivers noted that they have no idea if any services are available and would find a list useful. Most neurologists are affiliated with hospitals, which have on-staff medical social workers, who could connect caregivers with agencies. Understanding the stress that caregivers of highly dependent patients are under may influence scheduling in order to reduce the time patients spend in waiting rooms, another need mentioned.

The number one need stated by respondents in this study was respite care. Some even elaborated that they did not want babysitting services but trained professionals who were not afraid of epilepsy. Several caregivers explained that although there were a few day programs available in their counties for intellectually disabled citizens, almost none accepted adults with epilepsy, presumably because of fear of seizures. Some caregivers reported not having seen a movie in years because there was no one with whom to leave their child.

The second most frequently cited need was a support group where caregivers in epilepsy could get together to talk and share experiences or resources. One respondent explained that even an online group would be helpful. Another frequently mentioned need was a website for caregivers that included links to access services and social agencies with help for families taking care of chronically ill relatives. As more research in affiliate stigma and caregiver burden in intractable epilepsy is conducted, more evidence will be available to inform grant proposals for services and implement policies to assist families.

Conclusion

Despite these limitations, this study extends the past literature by providing a conceptual framework for studying affiliate stigma in intractable epilepsy. It also differentiates the perception of stigma from the perception of burden. There are very few caregiver studies in epilepsy that examine both children and adults; most focus on one or the other. Studies of perceived epilepsy stigma tend to gather data qualitatively with interviews or focus groups or quantitatively as an aspect of caregiver burden. Caregiver burden is usually operationalized with QOL checklists looking at depression, anxiety, or changes in physical health. Specific data about caregiving activities are rarely gathered in a quantitative way and then are usually either the presence or absence of the stressor. In this study, individual caregiving items were measured across a stress continuum which is helpful in identifying specific needs. As well, using stigma as a predictor of perceived burden is unique, and this is the first study to predict total levels of burden with patient age.

Further research is needed in examining the intersection of affiliate stigma and caregiver burden. Results from this study and respondents' comments about their needs may be incorporated into grant proposals. Caregivers of other intellectually disabled individuals could benefit from similar services as well and this is an important first step in easing the burden many face.

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APPENDIX A

SURVEY INSTRUMENT

Caregiving for those with Intractable Epilepsy

The following questions pertain to you and your family member with intractable epilepsy.

1. Are you a primary caregiver to a family member with intractable epilepsy? (check one)
    - ☐ Yes
    - ☐ No

2. Is your family member enrolled in the Alabama CBD Study? (check one)
    - ☐ Yes
    - ☐ No
    - ☐ They were enrolled but are no longer in the study

3. How old is your family member today? (age in years)

    ______ years

4. What is your family member's gender? (check one)
    - ☐ Male
    - ☐ Female

5. Please answer the following: (check "yes" or "no" for each statement)

a.	Because of my family member's epilepsy, some people are uncomfortable with me.	☐ Yes	☐ No
b.	Because of my family member's epilepsy, some people would prefer to avoid me.	☐ Yes	☐ No
c.	Because of my family member's epilepsy, some people treat me like an inferior person.	☐ Yes	☐ No
d.	Because of my family member's epilepsy, some people are uncomfortable with them.	☐ Yes	☐ No
e.	Because of my family member's epilepsy, some people would prefer to avoid them.	☐ Yes	☐ No
f.	Because of my family member's, some people treat them like an inferior person.	☐ Yes	☐ No

6.

Please check the box that most closely matches how you feel about each statement.

0 = This statement does not apply to me
1 = This statement applies to me and I find it: not stressful
2 = This statement applies to me and I find it: stressful
3 = This statement applies to me and I find it: very stressful

		0	1	2	3
1.	I can feel helpless/not in control of the situation.				
2.	My emotional well-being suffers.				
3.	It can put a strain on family relationships.				
4.	I can’t relax because of worry about caring.				
5.	I don’t have enough private time for myself.				
6.	I can’t devote enough time to other family members.				
7.	I feel angry about the situation.				
8.	I feel guilty about the situation.				
9.	It is physically tiring.				
10.	The person I care for is immobile/has problems getting about.				
11.	The person I care for doesn’t always help as much as they could.				
12.	My physical health has suffered.				
13.	The person I care for needs a lot of help with personal care.				
14.	My sleep is affected.				
15.	The person I care for is incontinent.				
16.	It restricts my social life/outside interests.				
17.	I can’t have a break or take a holiday.				
18.	I can’t see friends as often as I’d like.				
19.	Professional workers don’t seem to appreciate the problems caregivers face.				
20.	I don’t get enough help from the health and social services.				
21.	It causes financial difficulties.				
22.	My standard of living has fallen.				
23.	Relatives don’t keep in touch as often as I’d like.				
24.	Some family members don’t help as much as they could.				
25.	The person I care for sometimes manipulates me.				
26.	The person I care for can demand too much of me.				
27.	I no longer have a meaningful relationship with the person I care for.				
28.	The person I care for doesn’t always appreciate what I do.				
29.	The behavior of the person I care for is a problem.				
30.	There is no satisfaction to be gained from caring				

7. What services do you feel would be most helpful to you and your family?

____

____

____

____

____

____

____

Background Information

Almost done now! In order for us to more fully understand people's responses to the previous questions, we are asking you a few things about your background. *Please remember that continuing this survey is voluntary. You may stop the survey at any time.*

8. What is your gender? (check one)

- ☐ Male
- ☐ Female

9. What is your age? (fill in the blank)

_______ years

10. What category best describes your race/ethnicity? (please choose up to 2)

- ☐ White
- ☐ Black or African American
- ☐ Hispanic or Latino
- ☐ Asian
- ☐ American Indian or Alaska Native
- ☐ Native Hawaiian or Other Pacific Islander

11. What is your marital status?
    - ☐ Never married or in a committed partnership
    - ☐ Married/In a Committed Partnership
    - ☐ Divorced
    - ☐ Widowed
    - ☐ Separated

12. What is the highest level of education that you have completed? (check one)
    - ☐ Did not graduate high school
    - ☐ High school graduate or GED
    - ☐ Vocational or Trade school graduate
    - ☐ Some college or Associates Degree (2 year degree)
    - ☐ Bachelor's Degree (4-year degree)
    - ☐ More than a 4-year degree

13. What is your annual household income level? (check one)
    - ☐ Less than $5,000 per year
    - ☐ $5,000 to $9,999 per year
    - ☐ $10,000 to $ $14,999 per year
    - ☐ $15,000 to $19,999 per year
    - ☐ $20,000 to $29,999 per year
    - ☐ $30,000 to $39,999 per year
    - ☐ $40,000 to $49,999 per year
    - ☐ $50,000 to $64,999 per year
    - ☐ $65,000 to $74,999 per year
    - ☐ $75,000 to $99,999 per year
    - ☐ $100,000 to $124,999 per year
    - ☐ $125,000 to $149,999 per year
    - ☐ $150,000 to $174,999 per year
    - ☐ $175,000 to $199,999 per year
    - ☐ $200,000 or more per year

14. How many adults live in your house?

    ________ adults

15. How many children live in your house?

    ________ children

16. How did you hear about this survey? (check one)

- ☐ Alabama CBD Study
- ☐ Flyer
- ☐ Family member's neurologist
- ☐ A friend told me about the survey
- ☐ Word of mouth
- ☐ Other _______________ (please specify)

17. Please use this space to comment on anything you'd like to add.

____

____

____

____

____

____

Thank you for your participation!

If you have any other comments or questions you would like to share with us, feel free to email the researcher, Barbara Hansen at barbarap@uab.edu. Your email will not be tied in any way to your responses to the survey.

APPENDIX B

INFORMED CONSENT APPROVAL

CONSENT FORM

TITLE OF RESEARCH:	Stigma and Caregiver Burden in Intractable Epilepsy
IRB PROTOCOL NO.:	X160401005
INVESTIGATOR:	Barbara Hansen, M.A.
SPONSOR:	N/A
SPONSOR PROTOCOL NO.:	N/A

UAB IRB
Date of Approval 6-6-16
Not Valid On 6-6-17

Purpose of the Research

We are asking you to take part in a research study. The purpose of this study is to examine the experiences of people who take care of loved ones with intractable epilepsy. As a primary caregiver, you have a unique perspective of these physical and emotional challenges. Currently, there are very few social programs in place to address the needs of caregivers. Results from this study may be used to identify needed services and inform policies that might best help families like yours. We plan to have up to 200 people complete the survey.

Explanation of Procedures

If you agree to participate in this research study, you will be asked to complete a questionnaire. Some questions will be demographic, such as age, race, and gender. This survey should take no longer than 20 minutes to complete and will ask you about your experiences with stigma and the difficulties experienced by caring for someone with intractable epilepsy.

Risks and Discomforts

You may feel emotional when answering the survey questions about the challenges of caregiving but these feelings, if any, will most likely be mild. As this study involves the use of your identifiable, personal information, there is a chance that a loss of confidentiality will occur. The researchers have procedures in place to lessen the possibility of this happening.

Benefits

You may not benefit directly from taking part in this study. However, this study may help us better understand special challenges caregivers face and could inform policies designed to assist families dealing with intractable epilepsy.

Confidentiality

Information obtained about you for this study will be kept confidential to the extent allowed by law. However, research information that identifies you may be shared with the UAB Institutional Review Board (IRB) and others who are responsible for ensuring compliance with laws and regulations related to research, including people on behalf of the Alabama CBD Study and the Office for Human Research Protections (OHRP). The information from the research may be published for scientific purposes; however, your identity will not be given out.

Voluntary Participation and Withdrawal

Whether or not you take part in this study is your choice. There will be no penalty if you decide not to participate and you are free to stop the survey at any time. Your choice to stop the survey will not affect your relationship with this institution.

Cost of Participation

There will be no cost to you for taking part in this study.

Payment for Participation in Research

You will not receive payment for participating in this research.

Questions

If you have any questions, concerns, or complaints about the survey or interview you may contact Barbara Hansen. She will be glad to answer any of your questions. She may be reached by phone at (615) 397-6224 or email at barbarap@uab.edu.

If you have questions about your rights as a research participant, or concerns or complaints about the research, you may contact the UAB Office of the IRB (OIRB) at (205) 934-3789 or toll free at 1-855-860-3789. Regular hours for the OIRB are 8:00 a.m. to 5:00 p.m. CT, Monday through Friday. You may also call this number in the event the research staff cannot be reached or you wish to talk to someone else.

Legal Rights

You are not waiving any of your legal rights by signing this informed consent document.

Signatures

Your signature below indicates that you have read (or been read) the information provided above and agree to participate in this study. You will receive a copy of this signed consent form.

Signature of Participant Date

Signature of Witness Date

Signature of Principal Investigator Date

APPENDIX C

INSTITUTIONAL REVIEW BOARD APPROVAL

UAB THE UNIVERSITY OF
ALABAMA AT BIRMINGHAM
Institutional Review Board for Human Use

Form 4: IRB Approval Form
Identification and Certification of Research
Projects Involving Human Subjects

UAB's Institutional Review Boards for Human Use (IRBs) have an approved Federalwide Assurance with the Office for Human Research Protections (OHRP). The Assurance number is FWA00005960 and it expires on January 24, 2017. The UAB IRBs are also in compliance with 21 CFR Parts 50 and 56.

Principal Investigator: HANSEN, BARBARA
Co-Investigator(s):
Protocol Number: **X160401005**
Protocol Title: *Stigma and Caregiver Burden in Intractable Epilepsy*

The IRB reviewed and approved the above named project on 6-6-16. The review was conducted in accordance with UAB's Assurance of Compliance approved by the Department of Health and Human Services. This Project will be subject to Annual continuing review as provided in that Assurance.

This project received EXPEDITED review.
IRB Approval Date: 6-6-16
Date IRB Approval Issued: 6-6-16
IRB Approval No Longer Valid On: 6-6-17

Expedited Reviewer
Member - Institutional Review Board
for Human Use (IRB)

Investigators please note:

The IRB approved consent form used in the study must contain the IRB approval date and expiration date.

IRB approval is given for one year unless otherwise noted. For projects subject to annual review research activities may not continue past the one year anniversary of the IRB approval date.

Any modifications in the study methodology, protocol and/or consent form must be submitted for review and approval to the IRB prior to implementation.

Adverse Events and/or unanticipated risks to subjects or others at UAB or other participating institutions must be reported promptly to the IRB.

APPENDIX D

RECRUITMENT FLYER

Are you the primary caregiver of a family member with intractable epilepsy?

The University of Alabama at Birmingham

Department of Medical Sociology,

is conducting a research study to examine experiences of caregivers who take care of a child or adult family member with **intractable epilepsy**.

This voluntary survey will take no more than 20 minutes of your time.

As a primary caregiver, you have a unique perspective of the physical and emotional challenges faced by this population. Currently, there are very few social programs in place to address the needs of caregivers. Results from this study may be used to identify needed services and inform policies that might best help families like yours. To qualify, you must be 18 years of age or older and be a primary caregiver of a family member with intractable epilepsy. There is no charge for participation.

Please call the following number for more information 615-397-6224

or email barbarap@uab.edu.