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Perceptions of Illness Severity in Adults with Focal Drug-resistant Epilepsy

A dissertation submitted in partial satisfaction of the requirements for the degree Doctor of Philosophy in Nursing

by

Sandra Ruth Dewar

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ABSTRACT OF THE DISSERTATION

Perceptions of Illness Severity in Adults with

Focal Drug-resistant Epilepsy

by

Sandra Ruth Dewar

Doctor of Philosophy in Nursing

University of California, Los Angeles, 2019

Professor Huibrie C. Pieters, Chair

Drug-resistant epilepsy (DRE) is characterized by fluctuating periods of remission and relapse during which it is easy for the voice of the patient to be overshadowed by a bio-medical focus. Since little is known about perceptions of illness severity in DRE, the purpose of this qualitative dissertation was to explore how a sample of participants perceived their epilepsy and how subjective perceptions of severity shaped the treatment journey. The rationale for the study was influenced by the assumption that the course of disease is impacted by what patients know and believe about the condition. A purposive sample was comprised of 35 participants with DRE of which 16 were undergoing in-patient evaluation for epilepsy surgery and 19 had signed a surgical consent. Constructivist grounded theory was used for data collection and analysis from which four analytic categories were developed: 1) “Epilepsy is an uphill thing”, 2) “It’s just not knowing”, 3) “Waving the white flag” and 4) “Battling with myself”. Our findings revealed that considerations of overall illness severity were arrived at after much inner debate centered around uncertainty, shared suffering and disease burden. Tensions and contradictions were expressed in relation to the impact of unpredictable seizures, yet illness severity was frequently

downplayed, and participants found severity was difficult to rate. Pride was taken in coping well, yet acceptance of the epilepsy may also have contributed to a degree of treatment inertia. While epilepsy care felt directionless and life itself was on hold, other medical conditions were thought to be worse than epilepsy. We suggest that severity is a concept that is foundational to shared decision-making in epilepsy, and necessary to the timely execution of therapeutic options. Therefore, study recommendations include the importance of open conversations about illness severity. Nurses specialized in epilepsy care are well positioned to assess and influence patient and family attitudes around severity. A long-term implication includes the development of a practical instrument to assess severity that expands upon existing tools and includes subjective perceptions of illness.

The dissertation of Sandra Ruth Dewar is approved.

Jerome Engel

Eunice Eunkyung Lee

MarySue V. Heilemann

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University of California, Los Angeles

2019

Dedication

This work is dedicated to my family who always believed I could, should and would.

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1981	Bachelor of Social Science (Nursing), University of QwaZulu, Natal, South Africa.
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CLINICAL EXPERIENCE

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HONORS AND SPECIAL AWARDS

2016	Robert Wood Johnson Foundation, Future of Nursing Scholar
2016	UCLA Neurosurgery Outstanding Clinical Staff Award
2016	Fellow of the American Epilepsy Society

PROFESSIONAL MEMBERSHIPS

1992- present	American Epilepsy Society (AES)
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2019-present	Advanced Practice Advisory Work Group of AES Contribution: *member.*
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Dewar, S., Moseley, B.D., & Eliashiv, D. (2013). Safety, efficacy, and life satisfaction following respective epilepsy surgery in older patients. American Epilepsy Society 67th Annual Meeting, Washington, DC., December 9th.

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Chapter 1: Introduction

The purpose of this constructivist grounded study was to explore the illness experience of a sample of participants living with focal drug-resistant epilepsy (DRE) to better understand how the severity of illness was framed. (A list of abbreviations used in this dissertation is provided in Appendix A.) Class 1 evidence has established the effectiveness of brain surgery for drug-resistant temporal lobe epilepsy (Engel et al., 2012; Wiebe, Blume, Girvin, Eliasziw, & Effectiveness Efficiency of Surgery for Temporal Lobe Epilepsy Study Group, 2001), but a major challenge in clinical epilepsy is the underutilization of resources, including surgical therapy that are available for DRE at specialized epilepsy centers around the country (Haneef, Stern, Dewar, & Engel, 2010; Jette, Sander, & Keezer, 2016). While the healthcare system itself may play a role in underutilization (Wiebe, 2016), we assert that a better understanding of patient perceptions of illness severity may help to address a vexing problem. Such subjective descriptions can be used in the development of strategies to promote collaborative and timely decisions between clinicians and patients. However, questions need to be answered around shared decision-making including how much involvement patients want, what they want to know and how best to present treatment options (Dewar & Pieters, 2015). Since nurses in all clinical settings provide care for people with DRE, these practitioners are ideally placed to acknowledge illness severity and communicate the need for treatment urgency. Increasing numbers of community-based nurses are also well positioned to inform patients about severity and influence the trajectory of epilepsy care. Thus, disseminating research findings around the important topic of severity has the potential to raise awareness of contemporary challenges in DRE among patients, their families and health care practitioners to advance standards of epilepsy care.

Particularly relevant for this dissertation is that standardized measures of seizure severity have been developed for the purpose of conducting clinical trials, but how overall disease severity is represented has not been widely explored (Thurman et al., 2011). As a way to fill important knowledge gaps, the present study goes beyond the clinical details of seizure

severity to explore illness experiences that reflect perceptions of illness severity. For this qualitative study, the techniques of constructivist grounded theory methodology were used to reflect the meaning and processes of personal narratives (Charmaz, 2014). Our sample of 35 participants was purposively selected from a population of adults undergoing treatment for focal DRE at a specialized, level 4 epilepsy center, the highest level of specialization defined by the National Association of Epilepsy Centers.

Background and Context

Epilepsy is defined as “an enduring predisposition [of the brain] to generate epileptic seizures, and by its neurobiological, cognitive, psychosocial and social consequences” (Fisher et al., 2005, p. 470). Unfortunately, 30-40% of patients diagnosed with epilepsy do not respond to anti-seizure drugs (Tellez-Zenteno, Hernandez-Ronquillo, Buckley, Zahagun, & Rizvi, 2014). Drug-resistant epilepsy was formally defined by consensus agreement of the International League Against Epilepsy (ILAE) in 2009 as occurring when two well-tolerated and correctly prescribed drugs have failed to control seizures (Kwan et al., 2010). Much uncertainty is associated with the unpredictable clinical course of DRE making this one of the most serious problems in epilepsy (Tang, Hartz, & Bauer, 2017).

Epilepsy affects more than 3 million American adults (Tian, Boring, Kobau, Zack, & Croft, 2018) and contributes substantially to the national burden of chronic illness (Laxer et al., 2014). Although uncontrolled seizures impose substantial psychosocial and economic cost (Begley & Durgin, 2015; Kerr, 2012), many shortfalls are identified in the management of epilepsy that include the absence of practice protocols to guide the aggressiveness with which epilepsy is treated (Epilepsy Foundation, 2003). A published report by the American Epilepsy Foundation, “Living Well with Epilepsy 11”, highlights a general lack of public and professional awareness of the seriousness of epilepsy (Epilepsy Foundation, 2003).That this condition is

widely misunderstood and overlooked as a public health concern contributes to stigma and marginalization (De Boer, Mula, & Sander, 2008; Kerr, 2012).

The health care system itself may contribute to the underutilization of effective epilepsy therapy because of ignorance of modern approaches to treatment and ignorance around patient concerns (Keikelame & Swartz, 2016; Wiebe, 2016). Uncontrolled epilepsy is not benign and is associated with high morbidity and mortality. Rates of sudden unexpected death in people with uncontrolled epilepsy are five to 10 times higher than in the general population (Sperling, Barshow, Nei, & Asadi-Pooya, 2016).

While many clinical challenges exist around a chronic disorder for which poor health outcomes are noteworthy (Dalic & Cook, 2016), what people with epilepsy know and believe about the severity of disease may shed light on how DRE is experienced and how patients make decisions about seeking treatment. Illness perceptions play an important role in what people do about illness and whether behaviors are changed in favor of improved health outcomes (Stromback, Engstrom, & Walivaara, 2018). A condition such as epilepsy that does not have a static course creates clinical challenges (Berg et al., 2006; Choi et al., 2016). Many aspects of the natural history of DRE are not clearly understood by clinicians and patients including why and when seizures become refractory to treatment (Berg, 2004). Unfortunately, an unpredictable course of disease has been linked to lengthy delays in obtaining specialized epilepsy care thereby compounding disease burden (Berg, 2004).

The advantages of specialized epilepsy centers include the availability of a team of multidisciplinary experts and a range of therapeutic options (Engel, 2016). Although epilepsy surgery offers excellent and safe outcomes, only 2% of eligible patients undergo surgery, and they do so an average of 20 years after the onset of epilepsy (Berg, 2004). It was concluded in a recent review that a triad of fear of brain surgery, ignorance of the outcomes and willingness to tolerate ongoing seizures captures the reasons an effective surgical therapy is not embraced (Dewar & Pieters, 2015). It has been suggested that patients with long-standing epilepsy

become less empowered over time leading them to adopt a passive role in their care from which clinicians often take the lead (Varley, Delanty, Normand, & Fitzsimons, 2011).

The development of tools to measure illness severity constitutes an important challenge in clinical epilepsy and research (Speechley et al., 2008). Traditionally, the frequency of seizures has been relied upon as a measure of therapeutic effectiveness in new drug development and in trials of epilepsy surgery. However, seizure frequency is of limited benefit in outcome studies since frequency by itself ignores many aspects of illness experience that have the potential to influence quality of life (Cramer, Baker, & Jacoby, 2002; Shallcross et al., 2015). Seizure counts do not capture either the patients experience of illness (Borghs, Tomaszewski, Halling, & de la Loge, 2016) or the social implications of a seizure disorder for patients and caregivers (Thurman et al., 2011). The ILAE recently emphasized the importance of measuring overall disease severity in epidemiological studies in order to better understand and monitor the disease (Thurman et al., 2011). However, the ways in which seizure characteristics impact personal illness experiences and ultimately translate to perceptions of overall disease severity appear to be underexplored.

Current Gaps in Practice and Science

Shared decision-making is about incorporating into care what matters to patients. Therefore, unless patient perceptions of the severity of DRE are taken into account it may be difficult to address the timeliness of specialist interventions. By investigating this fundamental issue, new ways to address gaps in communication, education and advocacy may emerge. As priorities in health care shift towards holistic care, the voice of the patient is increasingly recognized as a central component of quality of care. Factors influencing treatment decision-making emerged as an overarching theme in the epilepsy literature to present a new perspective on the patient's role in determining the course of illness (Dewar & Pieters, 2015). How processes of treatment decision-making impact the illness trajectory in DRE have not been fully explained and represents an important epistemological gap. We propose that patient

perceptions of severity lie at the heart of this process in DRE to ultimately shape the timing of personal decisions about clinical care. The experience of living with epilepsy takes into account more than a bio-medical perspective. Whether and how people utilize medical knowledge as an effective resource rests on an understanding of cause and the expectations for cure (Scambler, 1994). A knowledge of patient beliefs and actions make it possible to draw attention to ways of overcoming health care barriers and promoting active patient participation in their medical care (Lee, Eun, Lee, & Nandy, 2012; Pieters, Iwaki, Vickrey, Mathern, & Baca, 2016). How to build communication skills in a population of patients often disadvantaged by memory and cognitive deficits is an important, yet untapped area of nursing practice and research. The tools and skills patients and families need in order to communicate priorities and limit uncertainty around a protracted course of DRE from the viewpoint of the patient have to our knowledge not been sufficiently explored.

Despite the efforts of government agencies, academic centers and national organizations to draw attention to advances in epilepsy and to encourage timely treatment, diagnostic and treatment delays remain pervasive (Epilepsy Foundation, 2003; Wiebe, 2016). If the public and professional image of a serious condition is to change, better ways are needed to communicate treatment urgency in DRE. A more complete understanding of patient perceptions of severity may lead to interventions that have the potential to improve long-term clinical outcomes and avoid disabling sequelae.

Therefore, the purpose of this of this study was to explore the illness experience of a sample of patients with drug-resistant focal epilepsy to better understand how the severity of illness was framed. The following three specific aims guided the research:

1) Explore perceptions of illness severity in the everyday lives of people with drug-resistant epilepsy in their own words,

2) Analyze and describe how perceptions of illness severity inform processes of treatment decision-making and actions around epilepsy, and

3) Develop a pragmatic, explanatory framework that encompasses the subjective components of illness severity.

Overview of the Study Design

A qualitative methodology was selected because human experience is a cornerstone of medical therapy and this approach offers scientific techniques for understanding personal perspectives. Illness narratives are richly captured in qualitative research and serve to highlight much that is not revealed in routine clinical encounters (Kleinman et al., 1995).

Constructivist grounded theory (CGT) is a rigorous methodology focused on exploring, analyzing describing and interpreting human action that has been widely applied in research related to illness experiences (Charmaz, 2014). A constructivist perspective provides a way to explore and understand the social reality of living with chronic illness and what individuals do about it (Charmaz, 2014). The underlying tenets of pragmatism and symbolic interactionism provided complimentary philosophical perspectives on how life with a serious neurological condition was lived and how individual views of illness severity shaped the trajectory of disease experience. Drawing upon what was useful and meaningful in personal accounts enabled us to construct a deeper understanding of the complex phenomenon of illness severity.

The current study was designed around a secondary analysis of a richly nuanced data set originally collected between September 2014 and March 2016. Data collection was based on 51 semi-structured interviews undertaken in a sample of 35 English speaking adults. Of this sample 16 were interviewed during a period of pre-surgical evaluation and 19 were interviewed shortly after an informed consent for surgery had been signed.

During the analysis phase of the research, we utilized CGT strategies that included three rounds of detailed coding, memo-writing and diagramming. Two researchers interacted with the data through the use of systematic coding techniques to identify categories, properties and dimensions. Using analytic techniques of constant comparison and analytic triangulation, we arrived at four core categories that served to structure the data. These techniques allowed us to

go beyond a descriptive level to reveal processes of meaning and action in the data. By means of abductive reasoning that builds upon inferences we arrived at an explanation of illness severity that provided a theoretical direction for understanding a dynamic and multifaceted concept.

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Chapter 2: Literature Review

Introduction and Purpose

Based on a recent survey, prevalence rates of active epilepsy in the United States (US) have increased, with 60% of patients reporting active seizures in the 12 months prior to the survey (Tian, Boring, Kobau, Zack, & Croft, 2018). In an effort to improve seizure-free rates in the population, recent recommendations of the US Center for Disease Control and Prevention (CDC) were focused on self-management support that includes the avoidance of known seizure triggers, and adherence to anti-seizure drug therapy (Tian et al., 2018). These recommendations are not new, and echo reports of the Institute of Medicine (England, Liverman, Schultz, & Strawbridge, 2012) and the Living Well with Epilepsy (Epilepsy Foundation, 2003), but do not extend to more forceful measures around the recognition of the severity of disease. Notably, no data was collected concerning epilepsy severity in the CDC survey.

Evidence-based clinical guidelines for when and why to refer patients with DRE to specialized epilepsy centers have been published (Engel et al., 2003). Unfortunately, despite the guidelines a sentinel paper showed that patients continued to live with epilepsy for about 20 years before being referred for surgical consideration (Haneef, Stern, Dewar, & Engel, 2010). We suggest that patient perceptions of illness severity, the focus of this dissertation, play an under-recognized role in treatment decision-making that may impact the course of illness and contribute to these 20 years. Since little is known about how patients perceive the severity of their epilepsy, exploring this knowledge gap may help explain the 20-year delay. Thus, the purpose of this literature review was to evaluate how the phenomenon of severity has been explored in the literature and reflected as a component of the illness trajectory in people living with epilepsy.

Method

An extensive literature search was conducted using the data bases of PubMed and Psych INFO. The focus of the search was to identify studies tracing outcome trajectories in epilepsy in order to strengthen understanding of human responses to a disease characterized by an unpredictable and evolving course. Published research related to outcome trajectories were sought and included studies about the life-experience of adults and children with chronic, refractory epilepsy. The search included combinations of the following keywords: “epilepsy”, “chronic illness or chronic disease or quality of life”, “trajectory or illness trajectory or quality of life trajectory”, “patient perception”, “outcomes and prognosis”, “epilepsy and illness experience”, “seizure severity”, and “perceptions of epilepsy severity”. Search strings using combinations of terms were applied. Exclusion criteria included non-English papers, papers about non-epileptic seizures, and studies specific to patient responses to anti-seizure drugs. To expand the search, electronic retrieval was done after hand-searching the reference lists of the main articles to identify key literature and citations not identified in the search strings. Although our study is focused on adults living with epilepsy, only three studies were found that address patterns of illness in adults, the other seven concerned pediatrics (Appendix B). We have included the pediatric studies since these illness patterns may shape illness experience as children transition to adulthood.

This review begins with a brief overview of how trajectory analysis is situated in the literature on the clinical management of epilepsy. The first half of the review includes a definition of the concept of illness trajectory and its application to DRE. This is followed by a brief discussion of how the clinical the social courses of epilepsy intersect. In the second half of the review an analysis of the literature concerning illness severity is presented, including perceptions of severity and the available measurement instruments.

Results

Defining the Concept of Illness Trajectory

The patient trajectory of illness is a concept that describes the direction, course, and time scales associated with a chronic condition (Jacoby & Baker, 2008). The concept draws attention to how interventions may (or may not) affect a clinical course. The concept also refers to phases or processes of care that occur over time, and includes the effects on patients, families and clinicians (Pescosolido, 2014). Accounts of illness survival are typically pieced together as people look back on how a diagnosis and its treatment was experienced (Godfrey, Price, & Long, 2018). As science advances, the ultimate goal of healthcare trajectories is to build predictive models that simulate disease progression and guide the timing of interventions (Pinaire, Aze, Bringay, & Landais, 2017). A contemporary focus in epilepsy research seeks to identify those factors that have the potential to predict a clinical trajectory at key points in time (Ferro et al., 2013; Sajobi et al., 2017).

For the purpose of the current review, the illness trajectory will be defined as the temporal nature of illness experience, shaped by varying and evolving disease processes that include psychosocial responses to living with epilepsy. Thus, the illness trajectory is a way to provide structure to the patient's experience of living with epilepsy.

Trajectory Analyses in Epilepsy

An important characteristic of the focal epilepsies is the occurrence of cycles of remissions and relapses. These cycles create patient trajectories that fluctuate and are not marked by linear or continuous events (Jacoby & Baker, 2008; Jacoby, Lane, Marson, & Baker, 2011).

Important foundational work on trajectories of illness experience in epilepsy were described by Jacoby and Baker in two unique studies that showed the intersection between the clinical and social courses of this chronic condition (Jacoby & Baker, 2008; Jacoby et al., 2011).

In the first study the relationships between five different seizure frequency patterns and quality of life (QoL) were traced (Jacoby & Baker, 2008). The five clinical patterns included experiencing just a single seizure, a few seizures, intractable seizures, good seizure control after surgery and poor seizure control after surgery. As a measure of patient experience, QoL was associated with each seizure frequency pattern and represented by line diagrams that give visual shape to a series of outcomes. Key findings included overall poorer QoL in those with active epilepsy. Greater negative impact was associated with both seizure recency, the time of day they occurred, and whether seizures would be seen by others. Seizure frequency played a minor role as a QoL correlate, compared with the far greater influence of patient perceptions of seizure severity on QoL. In patients with disabling seizures, merely reducing seizure frequency was not sufficient to impact QoL. However, achieving seizure freedom after a surgical intervention had the potential to have a profound and favorable effect on QoL. These five patterns represented time scales in epilepsy that mimic the Chronic Illness Trajectory Model of Corbin and Strauss (Corbin, 1998), except that the epilepsy trajectory was more about learning to live with disruptive, and disabling illness than with a declining course of health leading to death as in the Corbin and Strauss model.

In a later study Jacoby et al. (2011) traced a series of clinical trajectories in 527 adults beginning with a first seizure. After a first seizure QoL profiles were recorded at baseline, 24 and 48 months. Five outcome groups were described namely, the occurrence of a single seizure only, early remission, late remission, relapse after a period of seizure-freedom, and those with persistent seizures (Jacoby et al., 2011). Subtle cognitive compromise when seizures began posed an increased risk for developing refractory epilepsy. For those with persistent seizures, disruptive effects on personal identity and social roles, and overall worse QoL scores in the domains of seizure worry, anxiety, depression and self-esteem were described (Jacoby et al., 2011).

The most extensive work contributing to an understanding of the trajectory of recovery after epilepsy surgery has been published out of the University of Melbourne, Australia. In a series of publications, the processes of adjustment needed to achieve a new state of wellness after surgery have been described by Wilson and colleagues (Wilson, Bladin, Saling, McIntosh, & Lawrence, 2001; Wilson, Bladin, Saling, & Pattison, 2005). Becoming seizure free encompassed psychological challenges conceptualized as the “burden of normality” and served to link personal perceptions of treatment success including personal views of cure and normality, making up for lost time, and discarding the sick role (Wilson, Bladin, & Saling, 2004, p. 13).

In a longitudinal prospective study the course of recovery was recorded every three months for 24 months in 89 patients after anterior temporal lobe resections for DRE (Wilson et al., 2005). Using statistical tools specific to response profile analysis (Boolean Approximation by Simulated Annealing-BASA), behavioral features were clustered for each time period enabling two distinct groups to emerge: good (58%) versus poor adjustment (31%). The remaining patients (11%) did not exhibit adjustment difficulties. Paradoxically, half of those falling into the poorly adjusted group were completely seizure free at 24 months. Outcome trajectories after surgery were heterogeneous, and multivariate features marked the longitudinal course of recovery (Wilson et al., 2005). The predictive value of this research included that adjustments were not immediate but occurred over time. The degree to which epilepsy was an established part of self-identity and the extent to which it had impacted the structure and function of the family was crucial to the process of patient recovery (Wilson, Bladin, & Saling, 2007). The transition from illness to wellness after epilepsy surgery was a process in which the synchronized expectations of patients and families were key. Transitions were most successful when the expectations of patients and families were matched regarding social and psychological adaptations around a seizure-free life (Wilson et al., 2007).

The temporal course of illness experience in adults with epilepsy appears to be understudied. Examples of ten studies published between 2010-2017 that address a time course in epilepsy are summarized in Appendix A. A few key findings are highlighted below.

Firstly, DRE is a condition in which outcome trajectories fluctuate and are not marked by linear or continuous events (Jacoby et al., 2011). Lack of knowledge about appropriate and timely interventions, have been cited as practice gaps that have contributed to a negative illness trajectory for many people (Engel, 2016; Erba, Moja, Beghi, Messina, & Pupillo, 2012b). Secondly, patients lacked confidence in remissions that occurred spontaneously or naturally and held views of greater future uncertainty compared to seizure-freedom accomplished via a deliberate intervention such as surgery (Micallef, Spooner, Harvey, Wrennall, & Wilson, 2010). Thirdly, uncertainty underscores how patients with DRE and the parents of children with DRE responded to a fluctuating course of symptoms (Jacoby & Baker, 2008; Micallef et al., 2010; Pieters, Iwaki, Vickrey, Mathern, & Baca, 2016). Uncertainty is a key observation reported in patient trajectories of cancer (Godfrey et al., 2018) and stroke (Hawkins et al., 2017), and may influence perceptions of illness severity and illness beliefs. Lastly, while interventions, such as epilepsy surgery for DRE, have the potential to alter the illness course, a new trajectory of wellness arises that carries challenges all of its own (Wilson et al., 2004).

Studies of the Clinical and Social Courses of Epilepsy

Outcome patterns hold important implications for understanding illness experiences, including how patients prioritize their medical care. Few studies have assessed the clinical characteristics that predict changes in QoL over time (Ferro et al., 2013). Patterns of experience related to new onset epilepsy and those that occur when disease becomes resistant to medications are discussed in the following section.

The clinical course: Outcome patterns in new onset epilepsy. Longitudinal trajectories in new-onset epilepsy in adults (Jacoby et al., 2011) and pediatrics (Berg & Rychlik, 2015) revealed similar, unpredictable seizure outcome profiles that complicate prognostic

estimates and carry significant implications for patient and family education. To address this gap, risk factors predicting patterns of poor QoL were prospectively studied in a cohort of 374 families caring for a child (aged 4-12 years) with new-onset epilepsy (Ferro et al., 2013). Baseline cognitive and behavior problems in children combined with parental depression were risk factors for poor QoL. When cognitive and behavioral problems, number of anti-seizure drugs, and family factors such as family function, stress and parental depression were taken into account, family factors had a greater influence on the child's QoL than the clinical characteristics of epilepsy itself (Ferro et al., 2013). Since it may be possible to modify family factors when family-centered health care is practiced this is a particularly important finding that was supported in later work by the same research group (Sajobi et al., 2017). These two studies provided evidence for recognizing that both clinical and social factors may forecast temporal changes in QoL to provide direction for appropriately targeted care (Ferro et al., 2013; Sajobi et al., 2017).

The clinical course: Outcome patterns in DRE. Two longitudinal studies were identified in the adult literature that described patterns of seizure relapse and remission, and the associated clinical predictors in patients undergoing medical management (Choi et al., 2016; Neligan, Bell, Sander, & Shorvon, 2011). In the earlier study, three outcome groups were modelled for 40% of patients that were not seizure-free five years after disease onset (N=290). The first group achieved sustained remission (10%), the second group never achieved any periods of remission (20%) and the third experienced intermittent periods of control (10%). Except for the number of anti-seizure drugs prescribed, clinical and demographic variables including gender, age, age at onset, epilepsy type, and seizure type were not predictive of seizure patterns over time. Fewer anti-seizure drugs may predict better outcomes, however temporal patterns were variable and difficult to predict (Neligan et al., 2011).

Using the International League Against Epilepsy definition of DRE, Choi et al. (2016) at the Columbia Comprehensive Epilepsy Center performed a retrospective chart note review to

analyze seizure outcome patterns in medically refractory adults (N=403) who were beginning a third anti-seizure medicine. The mean age at seizure onset for the population was 24 years, with a mean duration of epilepsy of 18 years. Although longer observation time was a significant predictor of outcome ($p<0.001$), the clinical predictor most associated with seizure-related outcome trajectory was the type of epilepsy. After 65 months, outcome categories included subjects with ongoing seizures (53%), those with a complex, fluctuating course (16%), and those with prolonged seizure freedom (31%). Regardless of age of onset, patients with focal temporal lobe epilepsy and encephalopathic generalized epilepsies were less likely to achieve remission than other types including unclear and focal generalized epilepsies (Choi et al., 2016).

Authors of this ongoing longitudinal study suggested that 65 months is insufficient time to capture a complete understanding of seizure outcome trajectories. Long periods of waiting to achieve seizure control have obvious psychosocial impact many domains of independent living and create a vexing clinical dilemma. Clearly, a deeper understanding of the natural history of DRE in adults is needed in order to guide patient expectations related to the course of disease, and for the timing of treatment decisions (Choi et al., 2016).

The psycho-social course. Illness beliefs, explored through qualitative methods, appear to be central to a spectrum of human responses in epilepsy that include how people manage active seizures and adjust their lives (Harden, Tonberg, Chin, McLellan, & Duncan, 2015; Keikelame & Swartz, 2016; Yennadiou & Wolverson, 2017). The social context of illness requires taking into account when seizures began and provides a platform to understand the shape of chronic illness over time. Illness beliefs have been shown to statistically mediate the relationship between depression and QoL (Shallcross et al., 2015). Whilst illness beliefs may be a useful target to improve QoL, it was unclear which aspects of perception should be targeted. This study contributed to evidence that psychosocial factors have greater impact on QoL than clinical variables, such as seizure frequency (Shallcross et al., 2015).

Overall, both how people identify with epilepsy and their perceptions of illness severity may play a role in treatment decisions that ultimately change the course of illness and life itself. These aspects appear not to have been studied in any depth.

Illness identity. Illness events disrupt daily life, and as a consequence, how illness is understood requires interpretations and explanations about oneself. While biographical consequences are contextualized as a distinct component of most illness trajectories (Castellanos, Barros, & Coelho, 2018), people with chronic epilepsy suffer unique challenges with respect to self-identity, social relationships and the expectations of others (Rawlings, Brown, Stone, & Reuber, 2017). The degree to which epilepsy was integral to personal identity in young adults (aged 18-40 years) was influenced by age at onset, and thought to affect treatment adherence and the urgency with which more effective treatment was sought, thus impacting the course of refractory epilepsy (Luyckx et al., 2018).

For people with chronic epilepsy, transitioning across the life span is especially complex and insufficiently researched. Epilepsy that begins in childhood has life-long effects that impact social and educational success in adulthood (Jacoby & Baker, 2008). Illness identity may be integral to transitional processes that either compromise or facilitate transitions (Luyckx et al., 2018). However, not all epilepsy is experienced in a negative way. Personal enrichment was reported by study participants who attributed enhanced maturity and good coping skills to a diagnosis of epilepsy. One young person said, “I thought epilepsy was going to complicate my life…. It happened for the best. The illness encouraged me to grow up and develop” (Admi & Shaham, 2007, p. 1181).

Results drawn from an interpretive phenomenological study of ten patients with a mean age of 71 years whose seizures began in their early 20’s showed that loss of independence was compounded by heightened safety concerns. A longstanding diagnosis was associated with not being able to lead a “normal” life and a core characteristic of the epilepsy was a sense of “loss of control over body and mind” (Yennadiou & Wolverson, 2017, p. 89).

On one hand, the struggle to live a “normal” life was pessimistically expressed across age groups (Elliott, Lach, & Smith, 2005; Luyckx et al., 2018; Yennadiou & Wolverson, 2017). On the other hand, an overall optimistic view of personal coping was recorded in the written accounts of life with epilepsy, in which participants were eager to communicate how “normal” their lives were (Rawlings et al., 2017). In these written accounts, patients placed greater emphasis on biographical disruption as a consequence of the social course of epilepsy, than the experience of clinical seizures (Rawlings et al., 2017). While epilepsy was accepted by some patients as part of life and integral to personhood, others abhorred epilepsy, viewing “it” as something separate from the self, and that does not belong (Rawlings et al., 2017, p. 67).

Perceptions of illness severity. The distinction between illness severity (epilepsy) and symptomatic severity (seizures) is often blurred in the epilepsy literature. Illness severity is a complex construct in clinical epilepsy that can be statistically determined from a constellation of variables, namely seizure-related disability, number of anti-seizure drugs, medication side-effects, achieving seizure freedom for one year, and the presence of depression and anxiety (Sajobi et al., 2015b). Seizure frequency is commonly used as an indicator of disease severity but it is not a comprehensive measure of QoL outcomes, and is a poor indicator of the overall severity of illness experience (Vickrey et al., 2000). Patient reports of seizure counts are notably inaccurate and infrequent seizures may be just as disabling as frequent events (Baker, Smith, Jacoby, Hayes, & Chadwick, 1998).

Regarding age, younger patients endorsed higher self-reported epilepsy severity and more anxiety while older patients experienced lower disability (Sajobi et al., 2015a). Suggested explanations for these findings were that older people with epilepsy develop coping strategies and positive self-care behaviors that correlate with lower anxiety and less self-reported disability (Sajobi et al., 2015a; Yennadiou & Wolverson, 2017). The timing of such a shift in the course of chronic epilepsy may be a reflection of symptom tolerance and disease resilience but appears not to have been widely explored in the literature. These findings further emphasize the

important role of psychosocial factors, versus clinical variables in how chronic epilepsy is experienced and treatment is sought.

Perceptions of risk related to seizures in particular, and disease as a whole, is an important dimension of how epilepsy is understood. Seizures identified as a risk to independent living were described as chronic and incurable, yet the risk of dying from seizures appeared to be underplayed in some studies (Saada, Wang, & Bautista, 2015; Yennadiou & Wolverson, 2017). It has been suggested that patients minimize personal risk and create emotional "brackets" around issues such as sudden unexpected death in epilepsy, as a way to cope with an illness that is characterized by clinical uncertainty (Harden et al., 2015, p. 237).

Many factors shape the psychosocial course of epilepsy including how patients view themselves in relation to a diagnosis of epilepsy and their expectations of seizure control. As conveyed in the qualitative literature reviewed, narratives of illness experiences offer meaning, and explain actions that contribute to understanding the coping strategies of patients.

Representations of Illness Severity in the Literature

Patient perceptions of illness severity is a broad social concept closely tied to the impact of seizures and drug side-effects on daily life, and how patients create meaning around their epilepsy (Andermann, 2000; Kleinman et al., 1995). In this section, a discussion on how illness severity is represented in the literature will be followed by a brief synopsis of instruments that measure severity in epilepsy.

Preliminary studies suggested that modifiable factors associated with perceived disability need to be identified in order to target practical medical interventions (Sajobi et al., 2015a). Furthermore, how epilepsy severity is determined and the consequences for treatment urgency versus symptom tolerance are also understudied. The burden of epilepsy for families and caregivers is widely acknowledged (Kerr, Nixon, & Angalakuditi, 2011; Thurman et al., 2011), but how burdensomeness informs the experience of epilepsy severity is unclear.

Illness beliefs play an important role in how sickness is experienced and acted upon, but only a small number of studies were found in the general medical literature in which subjective perceptions of disease severity were explored. Personal beliefs about the seriousness of disease have been shown to influence adjustment and symptom control in chronic illnesses such as multiple sclerosis (MS) (Jopson & Moss-Morris, 2003), recurrent myocardial infarction (MI) (Stromback, Engstrom, & Walivaara, 2018) and asthma (Bidad, Barnes, Griffiths, & Horne, 2018). Among patients with MS a positive sense of control and lower illness identity were strongly linked to lower illness-related distress and less disability (Jopson & Moss-Morris, 2003). Similar findings can be extrapolated from studies in adolescents and young adults with epilepsy. Illness severity scores in a Swedish sample aged 13-22 years were inversely correlated with self-concept and attitudes towards the illness (Raty, Soderfeldt, Larsson, & Larsson, 2004). Israeli youth aged 15 to 24 years minimized their epilepsy and denied that it bothered them (Admi & Shaham, 2007).

Perceptions of illness influenced the reasons patients did not engage with optimal therapy in studies of asthma (Bidad et al., 2018) and epilepsy (Admi & Shaham, 2007; Prus & Grant, 2010). Similarities in self-management are apparent in these two conditions. Study findings in both conditions revealed that symptoms were downplayed, and poor symptom control was tolerated as part of living with these illnesses (Admi & Shaham, 2007; Bidad et al., 2018). Several authors have highlighted the importance of developing strategies to better understand illness beliefs and ways to address misconceptions about treatments that impact outcomes in epilepsy (Bidad et al., 2018; Erba, Messina, Pupillo, Beghi, & Group, 2012a; Prus & Grant, 2010). Views of severity in chronic illnesses were not linked to a diagnostic label as shown in a recent experimental study (King, Harper, Young, Berry, & Voigt, 2018). Rather, judgments about disease severity were tied to thinking about disease impact on daily activities. Conclusions about severity were revealed as the consequences for valued aspects of daily life were realized.

Epilepsy-Related Severity Instruments

Several scales were developed before 2002 including the Liverpool Seizure Severity Scale (LSSS) (Baker et al., 1998), the VA Seizure Frequency and Severity Rating (Cramer, Smith, Mattson, Delgado Escueta, & Collins, 1983), the National Hospital Seizure Severity Scale (O'Donoghue, Duncan, & Sander, 1996), and the Seizure Severity Scale Questionnaire (SSQ) (Cramer, Baker, & Jacoby, 2002). These early instruments that are frequently used focus only on the clinical characteristics of seizures such as type, duration, frequency, seizure-related injury, and post-ictal recovery, however, do not take into account patient perceptions of the impact of seizures on daily life (Fisher, Nune, Roberts, & Cramer, 2015; Speechley et al., 2008).

A major criticism of these early scales is that content validity was not supported by evidence from qualitative studies (Borghs, Tomaszewski, Halling, & de la Loge, 2016). This limitation has led to questions about whether the items and domains in these instruments appropriately measure what patients think is important about severity (Borghs et al., 2016). To address this shortcoming, a conceptual model of seizure severity was proposed based on semi-structured interviews with epilepsy patients and clinical experts in epilepsy (Borghs et al., 2016). The authors identified 42 seizure related symptoms with 26 different impacts. The model revealed many complexities related to how illness severity might be measured. Perceptions of overall disease severity encompassed a combination of the personal and social impact of seizures that went beyond individual seizure events. The duration of seizures was an important component of seizure severity. In addition, differences were noted between how patients and physicians viewed the dimensions of clinical symptoms (Borghs et al., 2016). Patients focused on the experience of the seizure event including the warning signs and the post-ictal period, whereas physicians tended to focus just on the frequency of events.

More Recent Instruments: Global Summary Scores

The Personal Impact of Epilepsy Scale (PIES), developed in 2015, consists of four domains to track the overall impact of epilepsy (Fisher et al., 2015). Amongst the items in this

tool is a single global measure of QoL. While the impact of epilepsy can be determined by the PIES correlations with single global measures of severity and disability seem to be an important next step. However, the practical value of global ratings of disease severity in research and clinical settings in epilepsy remain to be more completely tested (Raty et al., 2004).

The first published single-item scale was the Global Severity of Epilepsy (GASE). This scale was developed and validated in 2008 as a reliable and easy way to assess the severity of epilepsy during routine pediatric clinic visits (Speechley et al., 2008). When tested with 250 adult patients, self-rated epilepsy severity was correlated with perceptions of disability (Sajobi et al., 2015a; Sajobi et al., 2015b). Another single-item scale, the Global Assessment of Disability (GAD), was developed in 2015 to assess disability in adults, but is not yet validated (Sajobi et al., 2015b). The GAD was shown to be an independent determinant of disease severity that mediated the effects of seizure freedom to significantly predict patient ratings of epilepsy severity ($p<0.05$)(Sajobi et al., 2015b). However, as with the GASE score, the GAD score failed to highlight the properties of patient experience, including how functional limitations such as driving, and employment status influence perceptions of disability and illness severity. Summary scores, while quick and easy to administer, may represent oversimplified ways to assess very complex issues.

This preliminary work on single global scores highlighted important differences between how patients and neurologists perceived severity. Patients focused on the impact of seizures on daily life whereas neurologists focused on frequency and severity of seizures, and overall stability of symptoms (Sajobi et al., 2015b). Uncertainty about treatment responses and the time it took to stabilize the disease changed the perception of epilepsy severity in the minds of physicians (Chan, Zou, Wiebe, & Speechley, 2015). In another study, post-ictal recovery time was reported by patients and care givers as the single most important factor when considering the severity of seizures, yet received less attention by physicians (Borghs et al., 2016).

Growing evidence supports the greater contribution of psychosocial factors over symptom variables in impacting quality of life in adults living with epilepsy (Borghs et al., 2016; Kerr et al., 2011; Shallcross et al., 2015; Viteva, 2014). Finding ways to explore the lived experience and beliefs about epilepsy in routine patient care may reveal perceptions of disease severity that explain the urgency with which patients view their epilepsy care and the actions they take. Determining the impact of illness has important ramifications for monitoring outcomes in chronic diseases such as epilepsy. In turn, the treatment decisions patients make can influence the shape and direction of a disease trajectory. Framing the social impact of uncontrolled seizures around the language of severity presents a new angle on the meaning of refractory epilepsy and its management.

Discussion

The purpose of this literature review was to explore how trajectories of illness are conceptualized in the epilepsy literature with a specific focus on the epistemology of severity among adults. To date, how the illness trajectory in epilepsy is conveyed has relied on comparisons of cross-sectional studies and a small number of longitudinal studies that encompass variable time courses. However, in patients with DRE, the chronic cycles of seizure remission and relapse create a sense of loss of control over the epilepsy and likely influence patient perceptions about the temporal course of severity. A body of literature was identified that makes conceptual distinctions between the biomedical course of epilepsy and the social course and reveals an intersection around QoL measurement.

Three central themes are apparent in the literature: (a) people who accept disabling epilepsy as long-lasting and integral to self-identity were less proactive in their care, and more likely to feel disempowered, (b) disruptions in biographical identity emerged as powerful determinants of the course illness in epilepsy, raising questions such as “who am I now” and/or

“who will I become?” and (c) developmental stages related to age at onset of epilepsy played a role in the perspective of severity around DRE.

An underestimation of illness severity lowers the urgency to treat refractory seizures (Keikelame & Swartz, 2016; Varley, Delanty, Normand, & Fitzsimons, 2011) and leads to deferred surgical decision-making that may prolong a trajectory of disability (Erba et al., 2012a). Arising as integral to the experience of DRE and fundamental to adjustment, are beliefs about causation and how deeply epilepsy is seen as integral to the self. Refractory epilepsy predicts identity roles (Luyckx et al., 2018) and also influences adjustment after surgery (Wilson et al., 2007).

Until 2008, it appears that most studies about the illness course were quantitative and did not report milestones that could be useful for predicting outcomes (Jacoby & Baker, 2008). Analyzing patient healthcare journeys enables barriers to effective treatment options to be targeted, so that advantage can be taken of “critical windows” for effective intervention (Baca, Pieters, Iwaki, Mathern, & Vickrey, 2015, p. 830). The narratives in clinical practice and in grounded theory research give meaning to human life, that paves the way to understanding how people with uncontrolled seizures move through time. Qualitative research compliments what we know from a clinical perspective and reflects the range of human experiences that impact how people chose to manage illness.

In conclusion, new treatment technology has the potential to alter the course of disease in epilepsy (Moshe, Perucca, Ryvlin, & Tomson, 2015), however, important components of timely treatment means care trajectories need to be formalized and a serious commitment from all stakeholders is required (Pinaire et al., 2017). Diagnostic and treatment gaps as well as gaps in education and advocacy have been reported as barriers to how disease burden is addressed (Sillanpaa & Schmidt, 2017). By drawing attention to the lived experience of severe epilepsy, awareness of the human, moral and ethical issues to be considered in clinical encounters were raised and goes beyond documentation of seizure frequency and drug side-effects. Unless

greater attention is paid to the social course of illness, encouraging patients to participate in shared decision-making and embrace more effective treatments may remain elusive.

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Chapter 3: Philosophical and Theoretical Underpinnings

The focus of this study is to explore, analyze and describe how people with epilepsy understand the severity of a disorder characterized by unpredictable, uncontrolled seizures. While all disease has a pathological course, it is the patients' perspective of illness that is integral to how a disease expresses itself (Charon, R., 2004). Human accounts of illness experiences take the form of unique, personal narratives set within a paradigm of body and self (Frank, 2013, p. 170). Although it is demanding for clinicians to enter the world of patients, that space that links the self and the body, it is through telling and listening that a mutual interpretation of illness experiences is reached (Charon, 2006, p. 102). It is through qualitative research that we strive to interpret and construct meaning derived from all that a participant might or might not disclose about the self.

The experience of an illness trajectory is a multidimensional phenomenon that includes how the person responds and adjusts to clinical patterns that change over time. Personal reality is constructed within the world an individual inhabits, and it is this personal perspective that shapes the understanding of experiences and what is believed to be true (Gergen, 2009). In the next section, a short description of grounded theory (GT) as the prelude to constructivist grounded theory (CGT) will be presented, followed by a discussion of the philosophical foundations informing CGT, namely pragmatism, symbolic interactionism and constructivism.

Constructivist Grounded Theory Methodology: A Brief Chronology

Grounded theory was proposed by Glaser and Strauss in 1967 as a systematic qualitative methodology for exploring, analyzing and interpreting what is happening in the context of an illness situation (Charmaz, 1990). What sets GT apart from other qualitative methods, is its distinct approach to data collection and analysis. Analysis in GT occurs simultaneously with data collection, and uses a strategy of constant comparison to distill categories in the data that can be differentiated from one another (Corbin & Strauss, 2015, p. 94). Analysis in GT involves a process of systematic and interpretive coding for the purpose of

constructing meaning. As theoretical categories develop, they are compared and refined, and serve to guide ongoing data collection (Charmaz, 2012).

The chronological development of GT reveals an evolving history that reflects the epistemological stance of the researcher (Charmaz, 1990). The development of GT in the 1960's and 1970's ultimately served to reinvigorate qualitative research as a legitimate methodology. Grounded theory methodology provided for a way to identify open-ended processes (as compared to static quantitative analysis) and challenged positivist assumptions that qualitative research was limited to a descriptive level of analysis (Charmaz, 1990).

Grounded theory was born from the merging of two schools of thought namely, Glaser's quantitative, structured and objective approach to data coding, with Strauss's symbolic interactionist perspective. In the late 1980's, Strauss's background as a pragmatist and symbolic interactionist led to his collaboration with Juliet Corbin, a leading scholar in GT methodology (Corbin & Strauss, 1991). As a result an interpretivist shift took place in GT that eventually moved the methodology away from the notions of passive discovery, to take into account that social worlds are actively constructed (Corbin & Strauss, 2015).

Constructivism is the philosophical position that what humans know is constructed from personal realities (Rodgers, 2005, p. 154). Human actions are not based on the way the world is, but how an individual sees it, and the meaning an individual attaches to actions (Gergen, 2009). Because there is no single reality, a constructivist approach examines process in order to uncover what is happening for an individual in the contextual situation of the individual rather than relying on assumptions of absolute, single reality.

As applied to this research, the term constructivism recognizes the interactive relationship between participant and researcher, and acknowledges the influence of the researcher in the process of constructing knowledge (Charmaz, 2014). Supported by her work on the impact of chronic illness on identity, Charmaz added constructivism to GT to focus on how people construct selves in the face of chronic illness, and live with "the practical struggles

of managing life" (Charmaz, 1990, p. 1170). Constructivist grounded theory (CGT) is shaped by the philosophical foundations of social constructionism influenced by the original ideas of Berger and Luckmann in *The Social Construction of Reality* (Berger & Luckmann, 1966). In more contemporary dialogue, Gergen expanded upon the constructionist ideas of Berger and Luckmann to enlighten scholarly explanations of the role of social relationships in how people construct their worlds. In other words, how we see the world and how we participate in it depends on social relationships (Gergen, 2009, p. 28).

As a research methodology, CGT goes beyond a descriptive level of inquiry, to include an understanding of the processes by which events occur, such that a window is opened on personal experiences that might otherwise remain hidden (Corbin & Strauss, 2015, p. 66). Interpretive processes centered on the co-construction of social reality contributed a new layer of vitality to traditional GT. Simultaneously, researchers are required to acknowledge their assumptions and their philosophical perspectives at the outset of a CGT study (Charmaz, 2014). Thus, upon embarking on this secondary analysis, the assumptions influencing our research are explained in Chapter 4.

Philosophical Foundations of Constructivist Grounded Theory

Grounded theory has philosophical roots in pragmatism, symbolic interactionism and constructivism; therefore, these are the philosophical underpinnings that influence and guide the approach of this research. A short description of each perspective follows, including how each applies to the study of illness experience in epilepsy. A summary of the key points of each philosophy are provided in Appendix C.

Pragmatism. Understanding what constitutes truth and knowledge has been fervently debated by philosophers (Magee, 1998). Pragmatism is a set of philosophical ideas that assesses the truth of beliefs or theories in terms of their practical usefulness (Moore & Bruder, 1999, p. 187). Mid-nineteenth century American pragmatists agreed that truth changes relative to the context of time, place and purpose, but each of the classic pragmatists held different

viewpoints on how people arrive at what they know and believe (Moore & Bruder, 1999, p. 187). C.S Pierce described knowledge as a tool for survival defined by what is useful and meaningful to society. William James took the debate one step further, to describe knowledge as a survival tool defined by what holds "cash value" for individuals (Magee, 1998, p. 186). For James, cash value is about arriving at what works best, or makes a difference from a subjective perspective (Popkin & Stroll, 1993, p. 276). Beliefs arise from actions that are repeated to become habits, and in this way beliefs are verified and become fixed notions (Menand, 2001, p. 356). From Dewey's view point, the consequence of thinking was not about finding truth, but about solving the practical problems of daily life, a form of pragmatism he called instrumentalism (Moore & Bruder, 1999, p. 188).

Pragmatists understand that what makes sense to people exists within personal experience and that rational thought from the person's perspective is the key to making decisions and solving problems (Menand, 2001, p. 351). Pragmatism serves to frame knowledge in terms of what is believed to be true, and what is believed to be the most practical for solving an individual's problems (Rodgers, 2005, p. 177). This group of thinkers challenged traditional philosophy, based on the notions that theories and ideas are not static, and that the world around us constantly changes dictated by what we notice. Pragmatism argues for a pluralistic reality based on the premise that there can be no single, fixed truth, but many different interpretations of existence (Charon, J., M., 2004, pp. 30-31). It is about the meaning of ideas as reflected in values, beliefs, or theories. Ideas, as truth, can only be investigated in terms of how useful they are, i.e. whether they hold cash value, and how they translate into meaningful human experiences (Moore & Bruder, 1999, pp. 187-190).

From the pragmatist perspective, there can be no grand *Truth* since human knowledge is tentative and changes. Further, beliefs do not reflect all of reality, but represent personal tools for coping and adapting. Pragmatist notions of truth are criticized for restricting views on human beliefs to what is practical or successful, to the exclusion of what is moral or correct (Popkin &

Stroll, 1993, pp. 274-282). Critics are of the opinion that it is only through a moral frame that we know what is successful, and desirable. To counter this criticism, pragmatists argue that the only way to investigate whether beliefs are true is through how they affect the human life of individuals (Popkin & Stroll, 1993).

Pragmatism applied to the study of people with epilepsy. How people with uncontrolled epilepsy act as they move along an illness continuum reflects adaptive strategies influenced by personal beliefs. Beliefs related to the cause of epilepsy (such as the belief that epilepsy is due to demon possession, is deserved or contagious), provide insight into the complex social meanings of this illness and the ways people cope with disruptive seizures (Andermann, 2000). In some countries, perceptions about epilepsy lead patients and families to consult both traditional and biomedical healers (Good & Del Vecchio Good, 1994; Otte et al., 2013). Health care decisions are influenced by what people think is correct and what satisfies personal preferences (Menand, 2001, p. 351). In keeping with the pragmatic philosophy of Dewey, what is seen to work, and what is believed to make a difference, offers the greatest personal meaning (Menand, 2001, p. 362).

Symbolic interactionism. Relationships between people are the basis of human social life and it is through processes of interaction that individuals form opinions and make decisions (Gergen, 2009, pp. 2-5). Derived from American pragmatism, and in particular the works of George Herbert Mead (1863-1931) and Herbert Blumer (1900-1987), symbolic interactionism (SI) is a sociological perspective that offers a way to explain what is happening in the social lives of the individuals we study (Charon, J., M., 2004, p. 29). Symbolic interactionism is not a predictive theory, but a dynamic perspective that provides a way to understand what events and actions mean to individuals (Charmaz, 2014, pp. 262-265). Words, objects and actions are symbolic representations necessary for human expression (Charon, J., M., 2004). It is through symbols that individuals are socialized, and the means by which they possess the ability to reflect on the world, and to move to through time and space. An individual learns about the

world based on interpretive meanings attached to words, objects and actions, and in this way social life is essentially symbolic (Charon, J., M., 2004).

The self is a central concept in SI. The self refers to how humans as both actors and social objects express themselves in relation to themselves and to others (Charon, 2004). Thinking was conceptualized by Blumer (1969) “as a symbolic interaction with one’s self”, a continuous, active process that is central to how people respond to the world (Blumer, 1969, p. 16). Both Blumer and Charon emphasized the self or the “me” as the object of the actor’s action. The concept of the “me” is important because it is the “me” to which self-identity is ascribed. Identity may take on many forms depending on the social and situational context (Charon, J., M., 2004, p. 86). The uncertainty of illness and the loss of identity goals has implications for the self, including what Charmaz calls a “disrupted” self. In this way, recovery from illness is about restoring identity in a process of “regaining a valued self” (Charmaz, 2014, p. 227).

Symbolic interactionism is not simply about cause and effect. Instead the perspective assumes that people are active in shaping their worlds (Charon, J., M., 2004), and not merely passive recipients of cultural values (Charmaz, 2014, p. 270). What becomes meaningful is constructed through processes of shared social interactions that rely on symbolic ways to communicate including language, words, actions, gesture, and facial expression (Charmaz, 2014, p. 262).

Symbolic interactionism applied to the study of people with epilepsy*.* With respect to epilepsy, the symbolic and cultural experience of disease plays important roles in shaping decisions surrounding treatment (Kleinman et al., 1995). Symbolic views of the cause of epilepsy, that have negative connotations for example, “having inflamed veins in the head” or appearing “dead” during seizures, contributes to stigma with the potential to limit social and economic opportunity (Good & Del Vecchio Good, 1994, pp. 838-839). Both positive and negative aspects of illness experiences impact a sense of self. This makes it important for

clinicians to consider the elements of patient narratives when striving to understand how patients adjust to, and cope with illness (Hustvedt, 2013, p. 173).

The consequences of a chronic illness such as epilepsy, reflect the interaction between biological and social processes over the course of time, creating a local ontology of illness that includes what is most valued in everyday life (Kleinman et al., 1995). In addition to views of the self, language and commonly used metaphors are powerful symbols of the ways in which meaning is attached to actions and situations (Charon, J., M., 2004). Whilst labels serve to frame illness experiences, illness-related situations are understood by the names attached to them (Charmaz, 2014, p. 272). For example, commonly used language that conceptualizes a person with epilepsy as “an epileptic” negatively influences the expectations of others (Noble, Robinson, Snape, & Marson, 2017). Rather, the term “person with epilepsy” was preferred by patients in a recent study because it places personhood ahead of disability (Noble et al., 2017, p. 20)

In summary, SI asserts that human behavior is not simply a physical response to a situation, but takes into account a local social context, and includes thinking as an active process located in the present (Charon, J., M., 2004, p. 29). The self as a core concept of SI contributes to GT research by enabling narratives to be told in such a way that personal and social identity is revealed (Charmaz, 2014).

In the quest to create knowledge using GT, experiences are described, and theories are constructed by researchers and research participants together. As described in the next section, the constructivist perspective is inherently reflexive as the views of both are considered, and sense is made of what occurs in time and place (Corbin & Strauss, 2015).

Constructivism. For the constructivist, reality is created against a background of shared human experiences and practices in support of a relativist ontology that includes what is known, and a subjectivist epistemology that attests to how it is known (Guba & Lincoln, 1994, p. 105). The world we live in is constructed around what is useful, predictable and beneficial to personal

well-being (Gergen, 2009). In this way, knowledge is constructed by ongoing cognitive reflections and by intersubjective social processes that reflect how life is shared (Schwandt, 2007, p. 38). The assumptions of constructivism are expressed in Berger and Luckmann's classic work, *The Social Construction of Reality* (1966), as a circular relationship occurring around three core concepts: a) externalization, b) objectivation and c) internalization (Berger & Luckmann, 1966, p. 61). Within these three concepts, externalization suggests that individuals create their personal worlds through relationships that are formed for a reason. Individuals create their personal worlds through social interactions that are external to the self and go beyond the self. When relationships are formed, new bonds are created that include other people and social entities such as institutions. Objectivation refers to the elements of everyday life that exist separately from the individual but are accepted and taken for granted as normal parts of orderly life and include, structures, institutions, and language. The term internalization is applied to what is the conscious acceptance of outside elements as part of one's own reality. What's becomes a personal reality is created through negotiation and consensus and reflects the acceptance of social norms and values. Berger and Luckmann argued for a dialectic or circular relationship among the three core concepts in which each influences the other (Berger & Luckmann, 1966).

The world we live in is a mutually created social construct, in which reality is grounded in continuous processes of interaction and negotiation. These ideas are central to contemporary dialogue about how people see the world and build their lives (Gergen, 2009). Social practices gain meaning through language that facilitates the communication of shared understanding (Wallace & Wolf, 1999). To the constructionist, the use of language is the heart of social life and binds relationships in patterns of shared living. Words do more than describe, they play a functional role in relationships and in sustaining traditional social values, and in sustaining traditional social values (Gergen, 2009, p. 11).

Constructivism applied to the study of people with epilepsy. Constructivism is not about truth or belief systems, but provides a way to dialogue about multiple assumptions reflecting the way life is (Gergen, 2009, p. 5). A central assumption for the constructivist, is that we understand the world as a result of living in it, determined by how we assume life is. Hence, a qualitative study that explores how people make sense of the severity of uncontrolled seizures and what they describe they do about an unpredictable clinical course in refractory epilepsy has relevance. Since constructivism calls for a recognition of each person's reality, constructivist inquiry invokes critical reflexivity on the part of the researcher; this requires, questioning fundamental truths and raising to consciousness that which we take for granted (Gergen, 2009, p. 13). Implicit in constructivism is the assumption of mutual interpretation and construction of meaning between researcher and research participants. The primary outcome is not to be able to present generalizable findings, but to plausibly represent the lived experience of those participating in the study (Appleton & King, 2002).

Conclusion

Philosophical considerations shape the methodology of qualitative research and provide a framework to explore and explain complex social dynamics. Philosophical considerations provide a directional compass for methodological processes necessary for conducting robust qualitative research (Appleton & King, 2002). As a scholar, I am challenged to foster interpretivist approaches to CGT that go beyond merely describing experiences, but also ignite fresh insights about how life is lived with a disruptive disease (Charmaz, 2014, p. 339).

Qualitative inquiry affords a meeting place for intellectual, political and social debate (Noble et al., 2017). The philosophical assumptions of pragmatism, symbolic interactionism and constructivism, are useful tools to broaden what we know of the social world, and how we arrive at understanding individuals' worlds (Schwandt, 2003). What is practical and makes sense to people as they seek improved health is that which is important and valued (Polifroni & Welch, 1999, pp. 467-470).

The literature about how people with refractory epilepsy experience the clinical and social trajectories of illness severity appears limited and reveals this to be an underexplored topic. Constructivism provides an opportunity to explore tensions and contradictions in illness experiences that may serve to compliment the body of quantitatively derived literature. The use of CGT as a methodology, offers a way to explore the efforts made by patients to cope with the fluctuating course of uncontrolled epilepsy. Many clinical and scientific advances have been made in epilepsy over the last 100 years, yet little is known of the complex human processes that influence how, why and when patients with refractory epilepsy make their health care decisions. As new therapies in epilepsy become available, knowing how patients make treatment-related choices becomes important to the effective communication of therapeutic options. Establishing what is meaningful and helpful to patients is the first step in patient-centered health care. Thus, CGT methodology is fitting for this dissertation.

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Chapter 4: Research Design and Methods

Introduction

The purpose of this constructivist grounded study was to explore the illness experience of a sample of patients with drug-resistant focal epilepsy (DRE) to better understand how the severity of illness was framed. The current research was a secondary analysis of a richly nuanced data set comprising 51 interviews with 35 participants completed between September 2014 and March 2016. The data set concerned a sample of adults undergoing treatment for focal DRE at a tertiary, specialized epilepsy center. The original data were collected for the overarching purpose of understanding decision-making processes specific to perceptions of illness-related risks and benefits in the sample.

The current research was influenced by the notion that what patients know and believe about a medical condition, determines how and when treatment options are pursued. Two sensitizing concepts guided our research that: (a) illness perceptions can be modified to positively change disease outcome (Neligan, Bell, Sander, & Shorvon, 2011; Shallcross et al., 2015), and (b) psychosocial factors have greater impact on quality of life in people with epilepsy than some clinical variables, including the frequency of seizures (Jacoby & Baker, 2008; Jacoby, Lane, Marson, & Baker, 2011). Thus, the aims of this grounded theory study were to:

1) Explore perceptions of illness severity in the everyday lives of people with drug-resistant epilepsy in their own words,

2) Analyze and describe how perceptions of illness severity inform processes of treatment decision-making and actions around epilepsy, and

3) Develop a pragmatic, explanatory framework that encompasses the subjective components of illness severity.

A qualitative versus quantitative methodology was selected for this research. Qualitative approaches look for meaning in the words of participants, and how they explain their actions rather than focusing on proving a hypothesis around cause and effect relationships (Hesse-

Biber, 2017, p. 5). A deeper knowledge of patient experiences around the course of epilepsy and patient views of illness severity may provide clinicians with fresh insight into ways to alter the trajectory of a serious, disabling disease.

Assumptions on Beginning the Study

At the outset of the project, it was important to be aware of my assumptions and biases. Keeping an open mind paves the way for new insights about that which is often assumed or taken-for-granted. When planning the original research, we assumed that what patients do about uncontrolled disease reflects a combination of how the natural history of epilepsy is understood and how the illness experience is viewed. Therefore, an analysis of illness experiences requires both tracing human responses to a pattern of symptoms and exploring disease in the context of social beliefs and expectations about the future.

The personal assumptions that guided this secondary analysis stem from my background in the social sciences, and my experience as a clinician in a leading tertiary epilepsy center. Having dedicated two decades of my professional life to the care of people with uncontrolled epilepsy, I naturally have personal biases and assumptions about how life with epilepsy is lived. A key challenge for professionals in epilepsy care is how to disrupt the lengthy and disabling course of DRE. Departing from this foundational challenge, three central assumptions guided this doctoral research: (a) patients and clinicians conceptualize DRE in different ways, (b) treatment decisions ultimately determine the course of disease but are deeply influenced by subjective patient perceptions about the condition, and (c) personal beliefs about epilepsy change over time to shape the way disease is incorporated into daily life.

With respect to specific aim #1 (perceptions of illness severity in everyday life), I believed that one of the biggest challenges to living with epilepsy is the uncertainty associated with cycles of seizure remissions and relapses. I have a hunch that phases and stages of care will be reflected in the participants' narratives to reveal a fluid and evolving illness trajectory. It is

however possible that perceptions of the course of treatment may not surface as patterns or phases to explain the way patients act over time.

With respect to specific aim #2 (how perceptions of illness severity inform decision-making about epilepsy), I assumed that epilepsy has an impact on self-identity, and that the condition is generally viewed as disabling. I also anticipated that time may change selves and relationships and that personal views of disability might be reflected in how our participants experienced the illness trajectory and what they did about it.

My assumption with respect to specific aim #3 (to develop an explanatory framework of dimensions of severity) was that people with refractory epilepsy 'live around' the disease and appear to accept and tolerate unpredictable seizures for many years. It was important for me to guard against the assumption that participants necessarily view their epilepsy in a negative light. The ways in which personal views of epilepsy motivate participants to seek care are likely to reflect a dynamic system including the views of supportive others, life events and aspects of the healthcare system.

Several strategies were employed to limit inherent bias. Collaborative discussion with the dissertation chair occurred at each step of the research design and throughout the analytic process. Towards the end of theoretical coding, the other members of the dissertation committee were also engaged in discussion. Since the dissertation chair and two committee members were not part of the clinical epilepsy team, they were less likely to be influenced by some of the same biases as the clinical members. The systematic analysis of CGT mandates critical engagement with small sections of the data at a time, and a process of constant checking of interpretations to enhance the rigor of the research. Dr Pieters and I met regularly to carefully compare interpretations of the data, and the conclusions drawn (Hesse-Biber, 2017, pp. 328-329). The strategy of reflexive memo-writing served to record our personal responses to the data, the meanings we gave to it, and how interpretations were linked to central concepts (Corbin & Strauss, 2015, p. 47).

The remainder of this chapter is focused on the rationale for selecting constructivist grounded theory as a methodology followed by descriptions of eight key methodologic components: (a) the research sample, (b) the recruitment process, (c) data collection, (d) data analysis, (e) supporting rigor and trustworthiness, (f) human subjects' considerations, (g) strengths and limitations and delimitations, and (h) dissemination strategy.

Rationale for a Qualitative Research Design

A qualitative methodology was selected because this form of inquiry is appropriate for research that is focused on how chronic illness is perceived relative to a changing clinical trajectory. As such, qualitative methodology compliments cross-sectional, biomedical research that tends to overlook the subjective experience of illness (Andermann, 2000; Kleinman et al., 1995). Since the personal meaning of chronic illness is constructed from situated subjective views, the perspective of the participant in terms of the self, as Corbin and Strauss state in their studies, may powerfully influence the clinical and psychosocial outcomes of refractory epilepsy (Corbin & Strauss, 2015). Patient narratives may change as life-events unfold, but it is through personal, descriptive accounts that the researcher can begin to understand what is meaningful from the perspectives of patients. In fulfilling its primary iterative purpose, the aim of qualitative research is to explore, describe and explain (Hesse-Biber, 2017, pp. 15-17). A qualitative approach provides a collaborative opportunity for researchers and participants to build a theoretical framework that captures the temporal perspective of illness experience and represents the viewpoints of the sample (Rapport, Clement, Doel, & Hutchings, 2015).
The objective of our theoretical framework was to locate an argument derived from the sensitizing concepts identified above, and that builds upon the analysis of theoretical codes (Charmaz, 2014, p. 311).

Rationale for Constructivist Grounded Theory Methodology

Grounded theory has direct application to research questions that seek to understand personal perspectives on chronic disease and that take into account how transitional processes

are managed over time (Morse, 2016, p. 85). In a chronic illness such as epilepsy, patient views of the disorder and its treatment may change as time goes by, to reveal new day-to-day challenges. To mirror the perspective of Charmaz, how people with refractory epilepsy "situate themselves in time" may serve to answer the question of why and how the lengthy course of uncontrolled illness is managed (Charmaz, 2012, p. 11).

Methodologically, GT serves to explore and explain social actions that do not exist in a vacuum, but instead are situated in personal settings (Charmaz, 2014, p. 234). By means of identifying patterns in social responses to DRE, our findings highlighted what it meant to live with uncontrolled epilepsy, and helped explain the actions taken by our participants.

The interpretivist position of the researcher is what ultimately paves the way to a social constructionist ontology (Charmaz, 2014, p. 235). As constructionists, we build upon the pragmatic and interactionist foundations of traditional GT(Charmaz, 2014). Constructivist grounded theory (CGT) is focused on the ways that a researcher gains entry into a participant's world to co-construct what is meaningful in the lives of participants. Constructionism reflects concerns that go beyond descriptions of behavior to include what motivates the actions of participants and influences the consequences of actions. From the outset of our project we remained true to personal reflexivity, since we know that what we bring to the research in the form of our own experience and personal biases influences how we see the data (Charmaz, 2014, p. 27). Since the participant's world is framed within a cultural and social context, how the researcher interprets that context plays a role in how meaning is eventually constructed (Charmaz, 2012).

The Original Study

With reference to the PhD Handbook of the School of Nursing, this proposed secondary analysis was approved by the committee chair as it draws on original data, develops new research questions, and is based on a solid conceptual framework that reflects understanding of the qualities and limitations of the data set. Deeper analysis of the unexplored data was

expected to address a gap in knowledge by shedding light on how patients who have endured many years of uncontrolled seizures describe the clinical and social factors that shape personal illness trajectories, to address a gap in knowledge. Descriptions of the time scale and course of events associated with refractory epilepsy may complement the perspectives of existing quantitative studies that address psychosocial outcomes in epilepsy. The power of GT lies in analytic techniques and the goal of coding for processes, actions and meaning. This way of analyzing enables connections to be made that may reveal patterns in the temporal experience of DRE, that have not been highlighted in the literature so far. The ultimate goal of the study is to understand participant experience in order to identify clinical strategies that may facilitate earlier access to specialized epilepsy care with the potential to shorten the course of DRE and impact the burden of disease.

A. Research Sample

Following approval of the Institutional Review Board, a purposive sample was drawn from adult patients undergoing treatment for focal refractory epilepsy at a specialized, tertiary epilepsy center. Recruitment occurred between September 2014 and March 2016.

Selection criteria. Included were English speaking adults, with suspected or confirmed focal drug-resistant epilepsy, who were able to sign their own informed consents. Patients who had undergone epilepsy surgery in the past were excluded out of concern that previous brain surgery may change the treatment experience at the time of interview.

The data is drawn from interviews with a sample of patients at two important treatment times on the continuum of care: (a) during an in-patient evaluation that is part of the pre-surgical work-up (pre-surgical group, n=16), and (b) after the surgical consent for resective temporal lobe surgery had been signed, (post-surgical group, n=19). The time-points were selected for the original study because it was assumed that these points represented important decisional moments in epilepsy care that potentially challenged participants to reflect on the personal meaning of uncontrolled epilepsy.

B. Recruitment Process

Contextual setting. Recruitment took place in the setting of a tertiary, specialized epilepsy center, at a major academic institution in California. The center is accredited as a level 4 epilepsy center with the National Association of Epilepsy Centers (NAEC) of North America. To meet this highest level of specialization, a level 4 center needs to offer the services of a specialized multi-disciplinary team together with a comprehensive range of clinical services. These services include the evaluation and treatment of all types of seizure disorders and the provision of advanced, state-of-the-art neuro-surgical procedures. By means of in-patient video-EEG evaluation, lasting five to seven days, the behavioral and electrophysiological correlates of the patient's typical seizures are documented. This evaluation, together with a detailed illness history, brain imaging and neuropsychological testing, form the key components of a comprehensive pre-surgical work-up for potential resective brain surgery in carefully selected patients with focal epilepsies. Once the clinical work-up is complete, the medical team determines surgical candidacy depending on the concordance of components of the work-up. If surgically eligible, the patient is offered an out-patient consultation to meet with the neurosurgeon and the clinical nurse specialist (CNS) (Sandra Dewar). During this consultation the work-up is reviewed, treatment options are discussed and the opportunity to sign the informed surgical consent is presented.

Pre-surgical subgroup. On admission, potential participants were screened by the clinical team for inclusion in the study based on the epilepsy history and physical examination. These screening criteria included the description of behavioral seizures, a history of suspected focal epilepsy, and a determination that the patient was a likely surgical candidate. Once identified, and within 24 hours of admission, I met with prospective participants to invite participation, and to obtain an informed consent to take part in the study. Four potential participants did not want to be interviewed and declined to participate.

Post-consent subgroup. Eligible patients on the surgical waiting list were called and informed that a research study was being conducted and that they will be eligible to participate once they had met with the neurosurgeon and signed a consent for brain surgery. If they were willing, I obtained an in-person consent to participate in the study before the consultation with the neurosurgeon. After meeting with the surgeon and signing the surgical consent, a convenient time was determined for the research interview. Of the 19 participants in the post-consent subgroup, six chose to be interviewed immediately after the meeting with the surgeon. The remainder were interviewed before surgery, at a time convenient to the participant.

Sample size. Data saturation in qualitative research refers to the depth and richness of the data, and occurs when the properties of categories are fully articulated (Corbin & Strauss, 2015, p. 139). Thus, saturation influences the sample size. Data saturation in GT research is influenced by both the research objectives and the skills of the interviewer (Charmaz, 2014, p. 214). After the properties of each category was fully described and since no new categories or properties were apparent in our initially coded interviews, we determined that data saturation had been reached with respect to the original research objectives (Charmaz, 2014, pp. 213-214).

Although a set sample size is not prescribed in GT (Corbin & Strauss, 2015, p. 135), the size of our sample (N=35) is slightly larger than common estimates of between 20-30 participants (Mason, 2010, p. 3). Our sample size was arrived at in a methodical way that included subjects with diverse characteristics that filled out the data in terms of similarities and differences (Corbin & Strauss, 2015, p. 141). Our participants were representative of a range of ages, ethnic and gender groups, and covered a spectrum of cognitive skill levels. The technique of constant comparison was employed to build the original data set and to determine saturation. Constant comparison is an analytic process for comparing pieces of data in order to identify differences and similarities. Various techniques were used to identify properties within each category. Through constant comparison, concepts are formulated from categories and defined

by articulating their properties and dimensions (Corbin & Strauss, 2015, pp. 85-87). Common to all research where data is collected using interviews, a skilled interviewer utilizes expert listening skills that allow the participant to take the lead in expressing understanding of the phenomenon (Roulston, 2010, p. 16)

Sample characteristics. Consent to participate in the study included signing HIPPA paperwork that gives permission to access protected health information in the medical records such as demographic and salient clinical characteristics including the neurocognitive reports. Demographic data was collected from the chart notes and confirmed with the participant. These participant characteristics included personal demographics (age, marital status, education, employment, self-reported ethnicity), duration of refractory epilepsy, number of anti-seizure drugs, and emergency room visits due to seizures or seizure related injuries over the past 12 months. In addition, for the post-consent group, side of brain and lobe of surgical resection were recorded.

The majority of participants (57%) were female, and the mean age at interview was 35.6 years (range, 18-68 years). The sample demographics and clinical characteristics of both subgroups (N=35) are reported in Appendices D and E respectively.

C. Data Collection

Interviews are a method of data generation that are aligned with the theoretical perspective of the researchers. The main data source for our study were semi-structured, personal interviews. To reduce bias, the interviews were conducted by a single, experienced qualitative researcher who was not working in the medical center. The pre-surgical subgroup (n=16) was interviewed during an in-patient pre-surgical evaluation, between day two of the admission and the day of discharge. Participants were interviewed for an average of 77 minutes on each of two days so as to avoid participant fatigue. All participants completed both days of interviewing. The post-consent participants (n=19) were interviewed in the out-patient setting for an average of 100.5 minutes. The study was not designed to require participants to complete

both the pre-surgical and post-consent interviews, therefore different participants made up the two study groups.

An audit trail of field notes and reflexive memos was created after each interview to guide potential refinements to the interview guide for future interviews and to illustrate the inductive techniques of CGT. In this way, a powerful aspect of the bi-directional flow of information between researchers and participants was documented.

Semi-structured interview guides (SSIGs). The SSIGs consisted of broad questions that were prepared in advance and structured in such a way as to facilitate in-depth, thoughtful answers. Open ended questions allowed the participant to take the lead, and to express what was personally important about a specific topic (Rubin & Rubin, 2012, p. 31). In keeping with processes of constant comparison and reflexive analytic techniques, data collection and analysis occurred simultaneously. Through this process of constant comparison and regular discussion between the researchers, the interview guides were slightly modified as the research progressed based on what the participants identified as important during the interviews. Thus, the interview guide served not only as a tool, but formed part of the analytical process, that included an active partnership between the researcher and the participant (Rubin & Rubin, 2012, p. 31).

The interview questions were framed to invite reflection on the depth and detail of illness experiences. Depth implies exploring deepening layers of meaning focused on why events occurred. In addition, the details of how an event occurred, and its consequences were sought. (Rubin & Rubin, 2012, p. 103). To elicit details, follow-up questions facilitated clarification of main questions in the interview guide. These included questions that may not be clearly interpreted and understood by the participant, or to deal with responses that needed deeper exploration. With the aim to collect a rich data set, attention was drawn to what was personally meaningful, including social interactions, emotional responses and the symbolic use of words and metaphors (Corbin & Strauss, 2015, p. 39). Overall, participants were encouraged to reflect

on their personal understanding of epilepsy and its treatment over the years and to give clarifying examples from their everyday lives. Internal consistency in qualitative research is enhanced by exploring what is said or not said, listening for how tensions are weighed in the constructs of personal understanding, and following-up on inconsistencies in later questions (Roulston, 2010).

Different semi-structured interview guides (SSIGs) were prepared for each of the two subgroups. Although the two subgroups overlapped in areas such as the impact of seizures on daily life, there were also distinct purposes for each interview in the original study. The goal of the interview with the pre-surgical subgroup was to elicit how epilepsy was understood, how living with uncontrolled seizures was perceived and how illness risks and benefits were described. For the post-consent subgroup, the goal was to explore how epilepsy surgery was understood and the essential individual decisional processes involved in signing an informed surgical consent. Some examples of questions from the interview guides that relate to the secondary analysis are given in Appendix F.

Procedure. A total of 51 interviews were audio-recorded in a private space in either an in -or out-patient setting. Audio-interviews were transcribed verbatim by a professional transcription company, The Transcript Co-op (http://transcriptioncoop.com). The transcribed interviews were checked for accuracy and de-identified to provide an accurate record of the interview. All data and coded information, including pseudonyms were kept secured in a locked file cabinet and password protected electronic files. The original recordings have been destroyed.

D. Data Analysis

The systematic process of CGT research begins with initial coding. Line-by-line coding keeps the researcher close to the words of the participant for the purpose of describing what is happening in the data. Analysis of the initial codes paved the way to create focused codes. Analysis of focused codes led to the development of categories, properties and dimensions.

While analysis in CGT follows a systematic analytic process, researchers may also develop wider research questions as categories develop and concepts become clear (Charmaz, 2014). The data that were analyzed for this dissertation was comprised of a total of 51 interviews with 35 participants, field notes and memos, and the demographic and clinical characteristics of the sample. The analytic strategies of CGT included coding the data using constant comparative methods, writing analytic memos diagramming, and collaborating with the dissertation committee. Each strategy is separately described in the sections below.

Coding. Coding involved three steps. The first step, initial coding, was completed in the original study. The second step in the analysis, focused coding, entailed a higher level of coding. The third step involved theoretical coding. Throughout the analysis, the goal of coding was not to summarize data, but to identify processes, meanings and actions in the narratives (Charmaz, 2012).

Initial, line-by-line coding enabled us to interact closely with the data and to assign meaning to small portions of text at a time (Hesse-Biber, 2017, p. 315). This initial coding utilized gerunds, the noun form of verbs, to enable processes to be seen within individual narratives. Labels were created and applied to phrases and sentences in the data based on the action of the participant.

During focused coding, the second level of coding, the data was further sorted, synthesized and analyzed. Focused codes were another way to label the data that allowed condensed chunks of data to be raised to a level of abstraction so that the meaning of the data could be sorted under themes. By comparing initial codes across participants those that most frequently occurred in the data and the most important overall for the study were identified within and across interviews. Analysis of these groups of focused codes led us to recognize the major themes in the data which are called categories in grounded theory research. After extensive analysis of the data of each category, we developed properties within each category. When the properties were fully developed, we did theoretical coding to show relationships

between categories and integrate the analysis in a coherent way. The value of theoretical coding which occured later in the analytical process is that it contributes to the strength of a comprehensive analysis and helps builds an analytical framework (Charmaz, 2014, p. 150).

Various strategies that were used to explore the meaning and processes evident in the data included diagramming, identifying assumptions and taking opposite views, constant comparison and questioning “what is happening here?” (Corbin & Strauss, 2015, p. 90). Comparing codes and quotations on the same topics contributed to the direction the analysis took (Charmaz, 2014, p. 140). Constant comparison allowed for data to be grouped and conceptually labelled, while theoretical comparisons enabled concepts to be explored, and broken down into explanatory properties and dimensions, or characteristics of the concept. Contrasting individual characteristics within the data to identify similarities and differences helped explain the varied landscape of how people with refractory epilepsy viewed the illness trajectory. Identifying the relationships between categories ultimately builds a theory (Corbin & Strauss, 2015, p. 94).

ATLAS.ti was the software program used to organize the data (Muhr, 2017). Although the program was not developed for CGT methodology it was useful for initial coding and focused coding, and for highlighting the vivo codes. I developed my own system for differentiating the codes. I found it useful to record the analytical memos in ATLAS.ti, as this kept my thoughts together and helped to formulate the categories.

Memo-writing. Writing successive memos is an essential and indispensable analytic tool in CGT. Memos are written notes that reflect the researcher’s analytic ideas, hunches and questions (Charmaz, 2014, p. 162). Memos written early in the analysis to facilitate my interaction with the data and to help identify ‘what is happening’. Towards this goal, quoting the participants exact words, in the form of in vivo codes, helped guide the recognition of meaning and processes and supported our interpretations. Later in analysis, more advanced memos served to document how our categories were developed and served to locate them within a

well-reasoned argument. Memo-writing occurred at all stages of this CGT study, and helped to conceptualize the data, and to expedite the work (Hesse-Biber, 2017, p. 311).

Diagramming. Diagramming is a form of memoing. As an intrinsic part of CGT method, diagramming is a way to visually represent categories and properties as well as their relationships (Charmaz, 2014, p. 218). Diagramming was used throughout the data analysis to formalize a story-line that reflected key phenomena as described by participants. This also showed relationships between who or what formed various parts of the lived experiences of participants.

Collaboration with dissertation committee. A more accurate understanding of the data was facilitated by taking a reflexive stance on interpretations. For this reason, Dr. Pieters and I compared codes throughout the coding process. At the level of theoretical coding, relationships between categories were created as part of an inductive process in GT that enabled a cohesive, credible theory to be developed (Charmaz, 2014, p. 150). During theoretical coding we invited the perspective of the committee members to enhance interpretive sensitivity of the data.

E. Supporting Rigor and Trustworthiness

The quality of qualitative research depends on the development of full and rounded categories that provide clear explanations of the data. Rigor in grounded theory research is concerned with achieving validity, and being able to support interpretations that are trustworthy and credible (Hesse-Biber, 2017, p. 349). Throughout the analysis, trustworthiness was fostered by staying true to the orderliness of grounded theory methodology. Conscious efforts were made to protect neutrality and consistency (Appleton & King, 1997). Transparency and continuous reflexivity were practiced throughout the analysis to keep our personal ontological and epistemological beliefs in check. A conscious effort was made to remain objective and value-free (Hesse-Biber, 2017, p. 22).

The strategies used to enhance dependability included checking analytic insights at each step of data analysis to avoid interpretive issues. Dependability in qualitative research refers whether a convincing account is reliably told (Hesse-Biber, 2017, pp. 328-329). As a research team we compared opinions on codes that were independently arrived at, to ensure that the findings ultimately made sense. To further enhance dependability we shared memos and reflective thoughts related to interpretations and pattern-making (Hesse-Biber, 2017, p. 327). Credibility in GT relates to whether interpretations are valid, and how well the findings compare with established knowledge. Credibility is concerned with integrity, the impact of which extends from the individual participant to the context of the wider sample (Hesse-Biber, 2017, p. 326).

The purpose of our qualitative study was to understand a complex phenomenon from the perspective of a sample of participants. While, insights from this sample may be transferable to people with DRE in other settings, the limitations imposed by our study setting were kept in mind. Generalizability in qualitative research denotes analytic generalizability and refers to whether the data captures the experience of the participants. A sample may not be representative of all cases, but if core concepts are sufficiently broad they may have wider application than originally intended (Corbin & Strauss, 2015, p. 377). Although different to generalizability in quantitative research, transferability in qualitative research is “comparable in its power” (Hesse-Biber, 2017, p. 60).

F. Human Subjects’ Considerations

Permission to conduct a secondary analysis for this dissertation was obtained from the UCLA Institutional Review Board (IRB). Thus, data analysis was subsequent to IRB-approval. To be part of the original study, participants signed an informed consent, and were provided with a copy of the signed document. Participants were made aware that there would be no direct personal benefit for participation, and the risk of harm related to the interviews was rated low. For the pre-surgical group, two interviews were administered to prevent the potential for

participant fatigue. Participants in both subgroups received $50 for completing each interview. On a few occasions, some participants experienced brief dyscognitive seizures during the interviews. Quick return to baseline cognition, enabled the interviews to continue after a short pause. Care was taken that the interviews did not interfere with parts of the clinical evaluation. Great forethought was given to the nature of decisions participants were making regarding elective brain surgery. With this in mind, the emotional aspects of surgical decision-making were considered, and sensitivity was employed in how questions were phrased, and answers pursued.

Privacy and confidentiality. All interviews were conducted one-on-one in a private space either in the patient's hospital room or in a small conference room. During the consent process, participants were informed that the conversations were to be audio-recorded and transcribed. Participants were offered the opportunity to switch off the tape recorders if they wished to share thoughts that they considered highly sensitivity. Confidentiality was assured at every stage of the research process. All identifiers including participant names, family names and names of clinicians and institutions were removed. Anonymity was protected by using pseudonyms in the transcriptions. Broad descriptors were used when quoting the words of participants, and special care was taken during the reporting of findings not to disclose events or information that may be linked to an individual.

G. Strengths and Limitations

A strength of the qualitative methodology applied to the proposed study, is that the rigorous exploration of human experience facilitates the construction of subjective meaning surrounding challenging health related issues (Morse, 2016). A strength of our study is that the sample reflects diversity with respect to demographic, and clinical characteristics. However, a limitation of the diversity of the sample is that the number of participants who self-identified as either African American or Asian was relatively small. Thus, a limitation of the research is that the perspectives of these two groups are not adequately represented. In addition, recruitment

was limited to a purposive sample of patients undergoing treatment at a specialized epilepsy center in an academic medical center, located in a large metropolitan US city. Since resources at this center may be different from other urban or rural settings, the transferability of results may be limited. Presenting for treatment in a level 4 academic epilepsy center did influence patient perceptions of illness and treatment options.

H. Dissemination Strategy

The strategy for disseminating results will be guided by the goals of the study and the anticipated interests of the audiences. Since the study addresses the perspectives of patients, the findings will be of interest to professionals caring for people with epilepsy and the lay public. For the scientific audience, the study findings will be prepared in the form of manuscripts submitted to scholarly journals. Subsequent to publications, dissemination for lay audiences, commentaries and narratives will be prepared that stimulate individual interest and meet the needs of personal advocacy. In addition, professional groups such as Epilepsy.com may provide an informational outlet for research recommendations that guide patient self-management. Opportunities for platform presentations at scholarly meetings or professional and public advisory boards will be embraced.

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Chapter 5: Results

Introduction

The purpose of this grounded theory study was to explore how a sample of patients with drug resistant epilepsy (DRE) perceived the severity of their epilepsy and how these subjective perceptions shaped the treatment journey. We believe that drawing attention to perceptions of severity as a component of the lived experience may provide insight into key treatment challenges in the provision of epilepsy care. Such an understanding may guide interventions that have the potential to positively impact the trajectory of this chronic illness.

Drawing from analyses of 51 interviews, this chapter begins with an overview of the experience of living with refractory epilepsy. This creates the foundational context for a deeper exploration of how illness severity was perceived by our sample. Three other categories were developed that gave the work an analytic direction. These categories showcase a dynamic process that contributed to insights about how participants thought about the severity of their epilepsy and the actions they took in order to live with it. These categories include (1) living around unpredictable seizures, (2) having to surrender in order to survive and (3) weighing a personal battle at the center of illness experience. An iterative framework illustrates the relationships between these four core categories and properties synthesized from our data (Appendix G). This symbolic representation highlights the evolution of illness severity as a function of time and subjective meaning. The trajectory of illness experiences of our sample appeared to be fraught with internal debates and tensions that were present within the narratives.

"EPILEPSY IS AN UPHILL THING": THE LIVED EXPERIENCE

Living with a multifaceted, disruptive illness held symbolic and personal meaning to our participants. The collective thoughts of our participants exposed a trajectory of lived experience that started with receiving a diagnosis of epilepsy and extended to the point of realizing that

seizures had become unresponsive to anti-epilepsy drugs (AEDs). Our study represented a range of experiences across a population with diverse intellectual functions, duration of illness and age at onset. Across interviews, “light-bulb” moments of realization were expressed showing varied degrees of urgency to seek new treatment approaches. Understanding a diagnosis of epilepsy that was unexpected and unwanted presented a difficult challenge for participants. The process of becoming refractory was understood to be life-limiting and damaging to the brain. Realizing what it meant to receive care at a specialized epilepsy center involved the intervention of a knowledgeable team of clinicians, and the ultimate hope of stopping the seizures.

Understanding a Diagnosis of Epilepsy

Our participants’ diagnoses of epilepsy had turned life upside-down, and the struggle to cope with it was described as an “uphill thing”. Emotions associated with living with the disease centered around sadness, shock and disbelief. Seizures that occurred out of nowhere with no traceable cause were hard to explain and meant the diagnosis was often questioned. One person, with seizures that started in her late 20’s said she spent at least seven years in denial because it was impossible to believe that the “nightmare” of epilepsy was something she could ever experience.

The sudden onset of seizures and receiving a diagnosis of epilepsy were frequently incompatible with subjective perceptions of being in good health and having no family history of epilepsy. As most participants had not witnessed seizures in other people and were not conscious when they themselves were having a seizure, the diagnosis was often denied, and the seizures were ignored. Epilepsy was incomprehensible to participants; it was confusing and foreign to their sense of personal identity. Irrespective of the age at onset, epilepsy was frequently characterized as an unwanted condition that created fear, worry and sadness. For some, it was invisible and easily dismissed, but at the same time, it changed one’s whole life.

The futility of medication trials and the restrictive nature of the condition were described as “sad” as was the diagnostic label.

A young man explained his experience when seizures began at age 14:

> They just qualified [labelled] me. They just put me in a group with no further testing. So, they kind of said, "Oh. Well, you're an average epileptic. Therefore, you're just going to be on medication. You're just having seizures. That's too bad, so sad.”

Epilepsy was understood as a complex brain problem, perceived as hard to diagnose and hard to treat. The reasons “why that one spot in the brain short circuits” was expressed as a mystery and associated with an uncertain prognosis. Acceptance of the condition was most often framed around personal beliefs about the cause of epilepsy. Unlike heart disease and diabetes, which were viewed as preventable, the onset of epilepsy was seen as something over which one had no control. Participants compared it to diabetes and some forms of cancer, with the conclusion that no simple diagnostic blood test was available. For some participants, the absence of visible evidence for epilepsy, such as normal brain imaging, further contributed to confusion around the diagnosis. Believing epilepsy to be a condition of childhood that is present at birth, reinforced doubt about the diagnosis when seizures began in adulthood.

Differentiating between diagnostic terms, such as seizures versus epilepsy created confusion and reflected a degree of stigma. Medical language that was hard to understand meant that clinic visits were experienced as overwhelming and dependency on parents and spouses was reinforced. Participants found it was necessary to rely upon parents or a spouse in order to cope with anxiety related to having a poor memory and being unable to provide an accurate interim health report.

Epilepsy was more easily accepted when it was determined to be a God-given as it could not be such a bad thing if it was God’s will for one’s life. A person with a three-year history of epilepsy said:

> I'm Christian, and I believe in God. And so, when they started happening, I just figured …God's doing something to me for some other reason. So, that's how I chose to look at it. And I didn't think of it as being bad. I didn't get mad. I didn't fight back for any reason. It's just what happened. There's really nothing else I could do.

Epilepsy was easier to accept when blessings and positive life-lessons were attributed to living with the condition. Some participants determined that epilepsy had helped them to overcome shyness, and to learn patience and empathy. A valued sense of personal strength was achieved in coping well, having faith and "keeping going" despite the seizures. Nevertheless, the question of "why me?" was commonly asked and some reflected upon feelings of being singled out by God and labelled as different. A man in his forties questioned his life-long illness and asked: "What did I do to you (to God), what made you choose me?" The option of surgery was also determined by some to be led by God, a view that factored strongly in surgical decision-making.

Some key social challenges around the diagnosis of epilepsy were exemplified in the words of a participant whose seizures began at age 60:

> Epilepsy is very frightening for other people and not the person that has it since I've never experienced it [lapses of consciousness of which she was not aware]. But I think people have approached me differently, thinking that I do have epilepsy and that I'm not understanding, or they're frightened of me. And some of my friends, when we're having conversations, talk to my husband and not including me. And I think it's because they're thinking I have epilepsy and not just seizures.

The metaphor of a "fish out of water" was used by more than one participant to depict that seizures are a frightening and undignified event. One person explained: "You see somebody go into full-body convulsions. Their eyes roll back in their head, and they're like a fish out of water. That'd scare anybody that didn't understand why this person's doing it".

Becoming refractory. Life with uncontrolled epilepsy was likened to a roller-coaster ride and treatment experience was described as "a long and winding road." The journey of epilepsy

was characterized as a great struggle that was “terrible for the brain.” However, while suffering from epilepsy was painful and depressing at times, it was also accommodated and accepted, and not allowed to devastate one’s life. Longstanding epilepsy was viewed as a part of life, the only life known, and for some was integral to who one is. With respect to his longstanding diagnosis one person reflected a shared view when he said, “it's almost like that's who I am and that's how I go”.

Ineffective seizure control despite polypharmacy was difficult to understand and periods of seizure remission and recurrence were confusing. Cure was felt to be unlikely when seizures stopped for a period of time, only to begin again. As time passed, uncontrolled seizures came to be viewed as worsening. The daily struggle around what was called “brains that aren't working” meant having to cope with a sense of loss of control in many areas of life. Despite ongoing seizures participants frequently heard their physicians say, “Keep on doing what you are doing.” One man reflected on his visits to the pediatrician as a child and said in spite of his diagnosis, “they [doctors] didn’t see anything wrong with me…they’d just write down notes, ask me questions, and they’d leave, and that was it.” Later when he was an adult, this man had conversations with his physician about the possibility of stopping the seizures. The participant heard the doctor say:

> “Your seizures are not as bad as others. There's worse that I have seen. And you, you're the type of person that knows when he's going to have a seizure, knows when it's over, compared to other ones.” But I go, “I want to know more.” He goes, “No. That's the best thing that there is right now. There's nothing else.”

Dismissive encounters like this one were confusing to participants and frequently led to minimizing, disregarding or forgetting about the epilepsy. Regret was expressed for the years lost to a course of illness that was unpredictable, a course that demanded sustained effort to not give up on one’s health. Brain surgery was seen as a way to stop seizures, to move on with life

and to possibly achieve cure. In the case of the participant quoted above, temporal lobe surgery was eventually performed almost five decades after the epilepsy began.

An educator with a four-year history of epilepsy expressed guilt about her limited knowledge of the disease. At the beginning of the illness she actively questioned her care and did much research, but over time she described becoming “lackadaisical” and less interested. In the face of poor seizure control, she like many other participants in our sample, felt that her care was “stuck”. Across intellectual levels, as measured by the verbal comprehension index (Appendix D), participants described the importance of improving their personal knowledge of epilepsy, however many said they simply lacked the motivation to do it.

Taking responsibility for health care was viewed as a shared responsibility between patient and physician, but in order to work together trust in the physician was key. Disappointment in medical care was felt when a clear diagnosis was lacking for a long time. Encountering challenges at the onset of illness, firstly the lack of a clear diagnosis and secondly the lack of epilepsy care, were commonly expressed. One participant said:

> I went through a lot before I saw the doctors here at [level 4 center], so you've got to understand what I went through before I got to this point. So, I mean I had one tell me it was pseudo seizures. One told me it was stress. Like I said, one doctor told me I was crazy, used the word "crazy." One doctor told me I was there for drugs. I mean, I went through a whole lot of torment before I got to the point of [epilepsy specialist] telling me in 2008, "You do have epilepsy. You do have the seizure disorder. We're going to go at this point, and we're going to find out what's wrong with you." I mean, at that point, I think it was the first time I broke down and started crying. And I'm going to cry right now [laughs] because I mean I - it was the first time a doctor told me, "There's something wrong with you.” [chuckles]

A range of beliefs was apparent around notions of cure. Some participants believed there was no cure. Others were ambivalent about the likelihood of cure, but at the same time they

welcomed the possibility. For some who viewed epilepsy as a life-long condition, the focus shifted from cure to managing the epilepsy. In this context, epilepsy was viewed by both participants and clinicians as something that simply had to be dealt with. This sentiment was reflected in the words of someone with life-long epilepsy: “Ever since 1981…all doctors were telling me ‘(Name) you’re never going to get cured. You’re going to die from this epilepsy.’”

Receiving care at a specialized epilepsy center. When participants received a comprehensive evaluation at a Level-Four epilepsy center that confirmed the diagnosis of DRE, it reinforced that seizures were not a figment of imagination. Prior to specialist intervention, seizures were seen as unlikely to get better and to have further negative impact on the brain. Participants stated that they valued the committed, multidisciplinary team of experts at the Level Four center. The participants heard about treatment options which engendered hope for the future. For the subgroup of participants who were found eligible for surgery, signing a surgical consent represented symbolic validation that a serious health condition existed that could be successfully treated after many years of feeling “stuck”. The possibility of achieving seizure freedom after epilepsy surgery meant life might be better, healthier and easier. Worry would be reduced and life for everyone would be calmer.

Key Points

The experience of living with “the uphill thing” that symbolized DRE included multidimensional perspectives of illness experience framed around the social positions of participants and the age at seizure onset. Across our sample, life was on-hold and medical care was viewed as stagnant. The limitations of living with epilepsy represented past and present struggles and included a future that was uncertain. There was a common desire to be viewed as a normal person and not someone with a disability. The possibility of achieving seizure freedom after epilepsy surgery represented a beacon of hope for the future.

"IT'S JUST NOT KNOWING": LIVING AROUND UNPREDICTABLE SEIZURES

Participants described epilepsy as a complex brain condition that was not generally well understood. Specifically, seizures characterized by an altered sense of reality were difficult to explain. Participants lacked descriptive words to adequately depict their unusual feelings and uncharacteristic behaviors that occurred without warning when seizures happened. The absence of a seizure-trigger and the fear that episodes could occur at any moment was experienced as terrifying and restrictive. A father spoke of his gnawing fear that seizures might progress to convulsions. Due to this fear he limited his social activities and often missed out on family gatherings. As he struggled to rate the severity of his epilepsy, he determined illness severity to go beyond the physical seizures to include psychological ramifications. He felt that it was safer to simply stay home because, "It's just not knowing, because that means I don't know when it's a good or bad time to be somewhere."

Participants described how an inner dialogue took place in their minds around coming to terms with the impact of epilepsy and resisting personal identity as a disabled person. They described facing a moral conflict if they used resources for the disabled such as the Access transport system or if they applied for Social Security Insurance (SSI), despite not having an outwardly visible disability. Important tensions existed in relation to life-style restrictions, perceptions of disability and existential meaning. To participants life was hindered by seizures, but not necessarily limited; life felt restricted but not disabled.

Living around the epilepsy became necessary to daily survival. As this category was analyzed three subcategories emerged to provide a deeper understanding of a multi-layered illness experience that explained how it was that participants tolerated severe symptoms for many years. Firstly, seizures that were not obvious to the self or others constituted a symptom that was hidden from view on many levels. Secondly, living around episodic seizures required normalizing life and minimizing the impact seizures. Thirdly, living with caution and taking precautions were expressed as necessary adaptive strategies for daily living.

Hiding Seizures

Despite never having seen their own seizures, participants hid the condition because of fear, embarrassment, and stigma. Living around seizures included concealing milder events and even lying about the frequency to family and physicians. One of many coping strategies was to "compartmentalize" the epilepsy as was the term used by one young woman to bury, hide and deny a seizure disorder that she did not outgrow. As seizures followed her into adulthood, she suppressed their existence by referring to them only as "episodes of dizziness". Seizures were banished to a separate compartment of life and never discussed but they remained an ever-present part of herself.

Seizures represented an unwanted symbol of ill-health and disability. The importance of openly admitting to having epilepsy was expressed in similar ways by participants. There was a risk associated with keeping the epilepsy a secret described as falling into a figurative "ditch from which you could not surface". Disclosing the condition created a sense of relief and was likened to climbing out of a deep hole. Coming to terms with ongoing seizures meant finding ways to absolve oneself of blame for the condition.

Hidden within the self. Auras were expressed as deeply personal experiences that happened "in one's own head" and were hard to describe to others. Participants reflected upon the social impact of losing awareness and then being amnestic for the seizure event. The absence of an aura as a warning was considered to be the worst aspect of having epilepsy, together with never knowing whether an aura might or might not progress to loss of awareness.

Seizures perceived to be too subtle to be noticed by others were easily dismissed and ignored, while episodes of anxiety and panic (a legitimate component of some seizure types) left some participants to believe that others thought they were emotionally unstable, or what was described as "crazy". Coping was about "fighting off" the onset of a seizure, wandering away, avoiding others and isolating oneself. In an effort to hide seizures from the world many of our

participants became housebound and socially isolated. When describing her seizures one person in her early twenties said:

> I don't have the convulsions, but I still have reactions. And it makes me look very weird and obviously not normal. And that was part of me keeping it very secret, as well as the thoughts that run through my head when I have the seizure. They're very negative.

***Lacking awareness*.** In contrast to seizures preceded by an aura, some participants had a complete lack of awareness when experiencing seizures that were hidden from the self. These "intangible", unseen symptoms were considered to be different from the more easily appreciated symptoms in patients with, for example, cancer or infection. Of the experience of a seizure one person said, "It is as if you were never there. You can't verify what happened…. Your mind is blank. You're left in the dark." Another participant described the experience as, "It's the conscious moving out of reality into this very weird space, this weird unknown space." Lacking awareness meant that witnesses had to be relied upon to inform what just happened. Seizures that occurred without an aura made it easy to portray these seizure types as not bothersome, to "sweep the events under the rug", forget about them, ignore them and pretend they were not there. However, seizures that included sudden blacking-out followed by precipitous falls were frightening for everyone and thus were less easily ignored.

Epilepsy was often described by participants as "a painful condition" because of post-ictal headache, muscle aches, tongue bites and other injuries. While these symptoms confirmed that a seizure had occurred, participants found it curious that pain, a clearly palpable symptom occurred as a post ictal phenomenon yet was absent at the onset of a seizure.

Changing patterns. Changes in seizure patterns that occurred over time could not be ignored and contributed to a sense of treatment urgency. For our participants, these changes included seizures that became more noticeable due to increased frequency or changes in seizure behavior. Time-of-day of habitual seizures also mattered to participants and family members. Whilst daytime seizures were considered harder to live with because of their impact on social

life, nocturnal seizures were described as simpler and easier to live with because they were less noticeable. One person said, “If you never see it [a night-time seizure], you never know you have it, unless others tell you or you wake up on the floor.” Although some night-time seizures were less troublesome, and more easily ignored, those that were accompanied by falling out of bed could not be dismissed. When nocturnal seizures began to occur in the daytime, or previously unexperienced convulsive seizures occurred, new safety restrictions were imposed with greater impact on daily life.

The parents of a young adult participant were reported to “hate” his nocturnal seizures and his siblings were saddened by them, but he minimized their impact on his own life. He talked about not caring and said, “I know I have it [epilepsy], I don’t really care. I mean I got it. Who cares? I mean, yeah, it would be nice to get rid of it, but it doesn’t really impact your life-well, my life.” By ignoring the seizures, he thought that worry would be reduced for his family, and he would avoid being singled out as needing special accommodations. While he downplayed the impact of seizures, mostly for the sake of his parents, he could not ignore the disabling impact of severe post-ictal headache that lasted for several hours the following day.

Normalizing and Minimizing: “Throwing Caution to the Wind”

Key components of living around DRE included minimizing seizure episodes and emphasizing that a normal life was being lived. Several perspectives about what constitutes normality were reflected in our data. While some participants claimed they had always lived “normal” lives, others, especially those with who scored lower on the verbal comprehension index, expressed ambiguity about what constitutes such a life, and whether accomplishing a normal life represented a personal goal. In addition, other participants defined themselves as “sick” despite attempts to be “normal”. While a passive and accepting approach to seizures was taken by some participants, the practical life-lessons of living with a chronic illness reported by participants included active decisions such as living as if you do not have a problem and avoiding self-pity.

Two examples illustrate how participants of the same age normalized and minimized their seizures. One, who had not known life without seizures, determined that he coped by “throwing caution to the wind.” In his words, “Epilepsy teaches people to live each day and to enjoy the present because life is uncertain.” Another participant, with a comparatively short three-year history of uncontrolled seizures, suggested the best advice he could give to another person with recent onset epilepsy was not to worry, and “not to be sad or mad.” In his view nothing could be done about the epilepsy, and therefore, “You've just got to take it [epilepsy] in stride, be happy." When asked what he would say to the persons’ parents, he said,

> You could tell them not many people die… you could just tell them their child's going to be fine. There's nothing different about them. They're still them. They're still that same person you knew before. Well, with me at least, I haven't changed. None of my personality has changed.

Inconsistencies were noted in what participants said about their lives. On the one hand, life was lived normally but, in contrast, life was on hold and the future was uncertain. When asked for greater clarity one person explained that the duration of her epilepsy had played a role, and that over time she had adapted. In her mind, this adaptation was normal, but it also contributed to treatment delays and “made the epilepsy harder to fix”. Normalizing was viewed as part of the process of adapting to illness, part of which was not keeping the epilepsy a secret.

A condition that was not physically apparent meant others “see you as normal”. Unfortunately, this meant the impact of disabling restrictions was sometimes underestimated. Social Security Insurance (SSI) benefits were denied to a participant in her late 40’s because she appeared well, walked independently and did not use a wheelchair. For this reason, the agent considered her epilepsy to be “just not bad enough” to qualify for disability insurance. She left the SSI office in tears saying, “I’m sick. I can’t help the way I look.” Another participant who was in her early 20’s, was refused SSI because her clustered seizures had not been sufficiently documented in the medical records:

> When I went to the ER, because my mom took me, my main provider told the [ER] doctor… “What is she doing there? She doesn’t have to go every time she gets a seizure.” That made us leave the hospital…. and not go back.

While our sample of participants with DRE reflected a wide range of clinical characteristics, it appeared that considerations of illness severity were not uppermost in people’s minds leaving uncontrolled seizures to be tolerated for many years. When a seizure occurred, immediate fear was provoked in participants and in those who witnessed them, and life was forced to stop for the duration of the seizure. However, once the seizure was over, routine life quickly resumed as if the seizure had never happened. In a similar way, clinic visits drew much attention to the disorder, but participants admitted that once outside the clinician’s office, the focus on the epilepsy was lost. Routine life resumed and time passed.

DRE, a condition that evoked episodes of loss of awareness and amnesia imposed unique implications for the self and others. Our sample described how life with DRE was about learning to live around the challenges imposed by uncontrolled seizures. Living around many daily challenges included managing, adapting and simply pushing through. Seizures were frequently minimized, ignored or accepted in order for life to be lived. From a practical point of view, epilepsy created a need for dependency and reliance on others. Although much of daily life required the constant presence of others, the physical reality of experiencing seizures, and the task of coping with the condition were ultimately faced alone. As a young woman said: “in my opinion [epilepsy] is hard to understand if you don't go through it yourself. I appreciate [friends] immensely, but it's just something you have to kind of go through it yourself to be able to completely understand it.”

Taking Precautions

Coping with the threat of random episodes of loss of awareness required monitoring one’s self and taking precautions that included warning others. The constant threat of a seizure

impacted every step of life. Viewed in this light, epilepsy imposed unusual demands on daily life that required precautions to be taken for the present and the future. Caution was necessary around taken-for-granted tasks of daily living such a crossing a street. Participants acknowledged that precautions were necessary but at the same time one person reflected on a loss of personal control and said, “You’re being told how to live your life that’s not yours to live.”

Because of the need for precautions, epilepsy was viewed as a condition that should not be kept private but should be openly shared so that people would know how to react. When and how to disclose the epilepsy stood in contrast to the need to conceal the condition and was ultimately related to safety. As one youth said:

> Most of the time there isn't someone there. But, yeah. I know, okay, get somewhere safe, sit down, lie down. You know, just, I know what to do, I suppose. But if there is someone near me, I'll say, "You need to watch me right now and keep me safe. I'm going to have a seizure." Like I was saying, I had the episode on the escalator. I still have scars on my side from where I fell.

The properties of this sub-category included disclosing the condition, monitoring personal well-being and planning ahead.

Disclosing. While non-disclosure was a way to ignore the severity of symptoms, it was not always practical to keep the epilepsy a secret. Selective disclosure arose as a pragmatic component of daily life and a crucial strategy for managing the unpredictability of seizures. Informing others about the epilepsy included warning about possible seizures and telling people how to react. Disclosure was seen as an important part of considering the impact of seizures others and for the safety of self and witnesses. This was especially necessary when the risk of convulsions was high, or when seizures were followed by paranoia, aggressiveness and agitation. Participants relied greatly on friends and family irrespective of age. A mother described preparing and reassuring her four-year-old:

> If Mommy falls and I'm not answering and I may be shaking, don't worry. It's going to pass. Just, you know, pass your hand on my head or my shoulder and wait until Mommy calms down. She knows how to use the phone, that she can call Daddy. And just wait until I wake up, that she shouldn't worry, not get scared, that I'm going to be okay.

Social risk was involved in informing others and therefore decisions about when to tell and who to tell were selectively made on a "need to know" basis. Participants were aware that seizures provoked fear and embarrassment, so the condition was disclosed only to those who were likely to understand it, and once a trustworthy relationship had been established. Disclosing epilepsy was often met with surprise. Many participants echoed the sentiment expressed by one man who said, "Because [people] can't see it … and they kind of have a reaction as in, "Well, you look perfectly healthy." And my reaction to their reaction is kind of like, "I know."

Monitoring the self and planning ahead. Living with epilepsy across our sample was about embracing an outwardly normal interactive life and avoiding social isolation. Whilst it was important to avoid being controlled by the epilepsy, participants found it necessary to inwardly monitor how one feels. Constant mindfulness of impending seizures required a process of continually planning ahead and monitoring personal well-being that was described as living from minute-to-minute. To explain this process a participant with life-long epilepsy said:

> You have to have two streams of thought in your mind. One is, okay, just like everybody else, you are doing things. You are interacting. On the other hand, there is this parallel thing running, which is very introspective. You're always kind of taking a pulse of yourself. How am I feeling now? How is it going to go? Can I do this?

The time and energy required to monitor the self was likened to "keeping a car fine-tuned". To remain in good shape required staying positive, getting enough sleep, eating regular meals and coping with the limitations of having to use public transport. Planning ahead was about managing good days and bad days. Coping strategies included living with caution, avoiding

seizure triggers such as emotional highs and lows, and monitoring stress levels. While these strategies were described as effective, life lacked spontaneity.

Treatment decisions occurred in the context of reducing fear and worry. The futility of anti-seizure drugs and hearing that seizures are bad for the brain, propelled many participants to explore alternatives. Undergoing a comprehensive evaluation was undertaken as much for the self as for the benefit of others. Surgical decisions were frequently motivated by making life easier for others. While preparing for brain surgery was something so major that the epilepsy could no longer be kept secret, making a decision imposed another element of caution that required weighty discussions with friends and family.

Key Points

Unpredictable symptoms that encompass loss of awareness and amnesia represented a challenge that was unique to this chronic disease. Important tensions were observed around living cautiously, taking precautions or simply throwing caution to the wind. Disclosure was selective and undertaken with care but was part of normalizing the condition. Participants took pride in the coping strategies that had worked for them, and expressed the importance of remaining tenacious.

"WAVING THE WHITE FLAG": HAVING TO SURRENDER IN ORDER TO SURVIVE

The reality of living with epilepsy for these participants meant that having to rely on others and accepting family oversight was necessary to survival. Episodic loss of awareness impacted many aspects of independent living. In addition, seizures that impacted memory and slowed cognitive processing resulted in a changed self for some people. An old self was mourned and having to "surrender" to family care was about giving in to the sick role. The option of epilepsy surgery also represented conceding defeat to a condition that had been accepted

and lived around. When she signed the surgical consent one participant said, “It was almost like waving a white flag and saying, ‘I give up. This is it. You can take something out of me.’ "

The subcategory, missing self-sufficiency, contained two intertwined properties. The first property, suffering together, illustrated the awareness that both the participant and caring others experienced ongoing distress related to epilepsy. Uncontrolled epilepsy meant having to rely on and inconvenience others in order to accomplish practical tasks of daily life. Suffering together also entailed experiencing and witnessing seizures with implications for the self and others. The second property related to losing autonomy. Episodic, yet unpredictable seizures and an uncertain course of illness contributed to a sense of dependency from which many participants wanted to escape. Important tensions were experienced between these two properties. Others had to be relied upon but at the same time they were perceived as not always understanding the epilepsy. Mutual suffering was experienced around a condition that evoked loss of autonomy and led to concerns for burdensomeness.

Missing a Sense of Self-sufficiency

Unpredictable lapses of awareness created a self, a mind and a body that could not be relied upon. Personal views of self-sufficiency were negatively impacted when participants felt it was necessary to rely on others for everyday living. Seizures without an aura allowed for little or no time to provide a warning and meant family and friends were constantly on high alert. Care was needed when precipitous events were associated with falls and injuries, or when the post-ictal state included fear, paranoia and confusion. A 25-year-old recounted a dramatic episode about which he said, “I kind of blacked out and then woke up all wet. And, you know, I see my fish on the ground. I'm like, "What the hell happened?" [My mother] said, "You went through your fish tank. You got up and walked away."

While family members provided an essential lifeline in coping with episodes of loss of awareness, participants also felt that they were “at the mercy of others.” To accomplish everyday tasks others had to be relied on for physical help and vigilant surveillance. For

participants who had lived self-reliant lives before the onset of seizures, the transition to depending on spouses and children was traumatic. Handing over control of one's life represented a struggle for independence that was typical of daily life for our participants. Having to relinquish control imposed limitations on life that were sometimes viewed as a punishment symbolic of a life that was lived in handcuffs.

The threat of seizures at work was viewed as disruptive and had implications for obtaining and sustaining employment. Both losing employment and not being able to secure work signified further loss of control over the illness, and over life itself. Many participants stayed home because they did not drive, and the protection of family was needed.

To overcome social isolation took much strength. A young woman described having to learn to be independent of her parents, because initially "they never left my sight. Or I never left their sight." Although supportive friends were valued, participants said the everyday limitations of epilepsy were hard for other people to understand, including being alone in public, crossing a busy street, participating in sports or bathing without supervision. Particularly challenging for two divorced participants were the court-mandated, supervised visits with their children. Safety concerns related to post-ictal confusion and aggression meant there was little choice but to submit to the necessary oversight of family members.

Most participants yearned for independence and the freedoms of an epilepsy-free life. The impact of an illness that began in adulthood meant a reversal of prior independence and self-sufficiency. The need to rely on others for many aspects of day-to-day life was associated with disordered relationships. Feelings of inadequacy were fueled by dependency on friends and family. Not being able to drive or to work limited opportunities to contribute to the family. This inability to reciprocate in daily life was illustrated in the words of a young woman whose seizures began in early adolescence. She described feeling useless in society and likened herself to a "piece of trash". The inability to reciprocate much needed care led many participants to consider that they were a cause of worry and burden to their families. A common goal of

achieving seizure freedom was to be able to give back and to be a helper instead of the one needing help.

Suffering together. On several levels, the suffering of participants was mutual and intertwined with that of families and friends. Mutual suffering occurred around the traumatic experience of seizures that included seeing videos of one's own seizures and realizing the impact of seizures on those who witnessed them. The constant anticipation of seizures created mutual worry and anxiety that often changed family roles. Furthermore, seizures that resulted in amnesia disrupted valued memories.

Many examples were provided of mutual suffering related to uncontrolled seizures. One young man said, "My [siblings] are sad because I have lost so many memories." A 22-year-old said, "[When] you hear what your little sister has to say,…you understand that she's suffering…I also suffer because I know how they [the family] feel." Sadness was reflected around feeling socially isolated and deliberately left out. Participants were especially aware that peers and colleagues limited social contact because they were fearful of witnessing a seizure.

The embarrassment of seeing one's own recorded seizure was described as "torture". Waking up after a seizure to the concerned and tearful faces of witnesses was especially distressing. Personal suffering was foregrounded in the narrative of a young woman who explained what it was like to see her family looking down at her as she regained consciousness: "[As you go unconscious] you start seeing everything black. You wake up. You're in different clothes. You're in a different area. You see your family watching you like if you were a little pet in a like - [chuckles] little cage." Her words were symbolic of a diminished, confined and dependent self.

Constant worry about anticipated seizures, safety, and well-being created distress. Out of necessity, family roles changed, and illness became an essentially shared experience. About difficult days, the participant, a mother of teenagers said of her husband:

> He's had to put me in a wheelchair before because I couldn't walk after a seizure. I'm pretty sure we had a stroke after that one because I couldn't even walk. So yes, he's had to be physically lifting, physically caring. Um. He's my emotional support as well.

Because seizures created gaps in time, family members had to be relied upon to recount valued life events and to provide historical connections that would otherwise be lost to amnesia. One participant described the impact of seizures on his sense of reality and his need to rely on others to piece events together when he said, "I [gradually] start remembering small things of what happened. But I get to that point, it's like I don't know where it happened, but I know it happened… then you remember like people telling you… that's when you start connecting the dots little-by-little."

The presence of a family member at out-patient clinic visits to report the seizure behavior that participants themselves were unable to describe was viewed as necessary to help find treatment solutions. A young adult, with a 20-year seizure history, referred to his mother as his advocate, friend, protector and companion. Chronic seizures had impacted his ability to take initiative and he described needing his mother's help to navigate his schooling and his medical care. He especially valued his mother's presence during medical visits because, like other participants, his poor memory required that he rely on someone else to remember and report information, and to ask important questions.

The essentially shared nature of living with epilepsy was reported to be underappreciated by some clinicians who appeared to dismiss the crucial contributions of family members. An example of an out-patient visit was described by one man in his late forties:

> My wife was sitting with me, and she wanted to tell what I was experiencing and what she was seeing when I was having seizures. And he told her, "Are you the patient? Am I asking you the questions, or am I asking your husband the questions?" And I stood back, and I'm like, okay. And he goes, "I'm the doctor here. I ask the questions, and I ask

them to my patient, not to you. You're not the patient. So, don't tell me." So, I thought that was rude.

Losing autonomy. In order to cope with limitations in many dimensions of living, our participants reluctantly had to “wave the white flag” and submit to relying on others. The single most limiting aspect of losing autonomy was not being able to legally drive. The act of being able to drive a car and hold a license held social and symbolic meaning that was integral to personal autonomy. Participants of all ages viewed driving restrictions as a life-limiting obstacle that diminished personhood. Participants who had held a drivers’ license that was later revoked, reported feeling marginalized in society, as in “crippled” and “punished”. Family roles had to change and parenting obligations that included driving children to school could not be met. Having to depend on others for transport created an intertwined burden for self and others.

Depending on others for rides required living around the schedule of others and living at the pace of others. Having to wait for others created dependency and reinforced feelings of disability. While having to rely on others for transport became a necessary way of living, it came at the cost of living a spontaneous life. Spontaneity was considered to be a core component of normal living. This particular restriction represented a loss of personal dignity and was likened to be imprisoned. It was clear that the possibility of becoming seizure free and being able to drive represented strong motivation for undergoing epilepsy surgery.

Key Points

Seizures that robbed participants of consciousness at unpredictable times meant that minds and bodies could not be relied upon. Out of necessity living safely around epilepsy was about adapting to and submitting to varied levels of dependency on others. For most participants the investment and persistence of family members was key to moving care forward since coping with limitations could not be accomplished alone. The loss of autonomy in accomplishing the mundane and the grander things of life translated to a loss of spontaneity in daily life that was reflected as a core component of perceived disability in our sample.

"BATTLING WITH MYSELF": ADMITTING, RESISTING AND EXPERIENCING BURDEN

The terms admitting and resisting were used in this category to portray the internal struggle that culminated in perceptions of disease severity and treatment urgency. Life with seizures was viewed as full of obstacles and the first subcategory, admitting severity, was situated in the realization that there was urgency to better treat the epilepsy. This growing sense of urgency was reflected in words such as "I can no longer live this way". The second subcategory, resisting severity, addressed the seemingly contradictory idea that notions of severity were challenged and debated. Resisting the epilepsy and rationalizing that "epilepsy was not that bad" reflected an internal tug-of-war between admitting and resisting severity that happened over the course of the illness. This inner struggle portrayed as "battling with myself" summarized the futility of taking medicines that did not work. The third subcategory, experiencing burden, focused on the weighty experience of DRE. The tension between admitting severity and at the same time resisting severity lay at the heart of a burdensome battle. Experiencing burden was not apparent early on in the conversations but instead unfolded as the participants became more reflective.

Admitting Severity

As seizures became worse, memory loss progressed and quality of life worsened. Participants expressed concern about the worsening seizures that may have progressive impact on the brain. One participant justified the need for more definitive treatment saying, "My life is getting worse and worse. And having grand mal seizures while you are awake are horrible, horrible." Admitting severity encompassed having more bad days than good and running out of treatment options. A young mother spoke of resisting to admit the increasing severity of epilepsy for years by not taking the epilepsy seriously and skipping doses of medicine until she suffered a generalized tonic-clonic seizure. In hindsight, she determined that better advice from her clinician "would have saved me like four years of battling with myself and feeling that I wasn't in control of my life anymore."

Part of the inner battle was finding ways to cope with the embarrassment of seizures and to avoid the tendency to become isolated at home and “closed off from the world”. Several participants spoke of being “stuck” at home because they did not drive, and friends were afraid to spend time with them in case a seizure occurred. “Fighting” the illness included many struggles that were frequently underrecognized by clinicians and the family. A young woman in our sample was often homebound due to high seizure frequency and many injuries. She reflected on the emotional and physical severity of uncontrolled epilepsy in general saying, “In reality a lot of people are scared and trying really hard not to be in pain all the time.”

The impact of epilepsy on personal identity emerged as a key component of perceived severity irrespective of the age at seizure onset. Early-onset epilepsy was a familiar and integral part of life for one participant who referred to this as “the only life I know”. For others, epilepsy that began in adulthood was framed as involving of a major life change. For example, participants who once led independent lives saw that which was “taken away” because of seizures as personally diminishing. To this point, a participant whose epilepsy began in her mid-forties mourned a lost self when she said, “I like me, I don’t like this sick person.” Due to worsening seizures, memory loss and cognitive decline she had relinquished her physical, medical and financial responsibilities to her daughter. While the epilepsy limited her life, she reflected that it also took a serious toll on her daughter. At one point this participant contemplated suicide. A growing sense of urgency to get better, to be a better person and to regain an efficient and independent self, led her to consider epilepsy surgery.

Drug side-effects that were frequently reported to be worse than the seizures themselves also contributed to the increasing awareness of severity. For many participants, side-effects such as sedation were constant and consciously experienced. This was in contrast to episodic seizures for which many participants were often not conscious. One person in her early twenties reflected on the impact of anti-seizure drugs and said, “I have to live like an 80-year-old woman now. I have to sleep. I have to be on medication the rest of my life. I can't drink,

which was big in college, you know? My life has completely changed." Another young participant with a three-year history of seizures, reflected on drug side-effects that had her "coming off as an idiot" and necessitated urgency to be free of seizures.

Perceptions of an uncertain future also led participants to conclude that seeking better seizure control could no longer be postponed. A point of finality was reached in which life was likened to being up against a wall. Words such as "unsustainable", "hitting a road block" and "being at the end of one's tether" reflected implicit realization of the severity of illness. Because, for this sample, the seizures always came back after periods of remission, a normal life could not be lived, and the epilepsy was determined to be incurable.

The lack of urgency with which some clinicians approached seizure control was revealed through many of the narratives to characterize a core experience of DRE. The impression that ongoing seizures were to be expected and accepted as part of the pathology contributed to treatment inertia and left participants to conclude that care was stuck. Being simply "sick of seizures" and the high cost of epilepsy drugs reinforced the urgency to "fix" it. With much regret for lost time, and the distressing impact of uncontrolled epilepsy on his life, one person with a history of seizures longer than four decades said:

> Just like when the doctor asks you, "Are you getting seizures? Are the drugs helping?" And he says, "Okay, keep on doing what you are doing." I pretty much told myself the same thing. As long as something [does not go] seriously wrong, why upset the apple cart? Wish I had known where it would end up. I would definitely have done something about it. Looking back, I should have done something about it, but I don't know what.

Test results played a role for participants. Normal magnetic resonance imaging (MRI) findings created confusion around the cause of epilepsy and the treatment options. In contrast, a visible lesion on MRI provided tangible evidence that made it possible for participants to "see what I feel" and proved that the illness was not a figment of one's imagination or one's own fault. A visible lesion was especially helpful when words to portray aura sensations were lacking. One

person explained the meaning of seeing supportive evidence for epilepsy that suddenly began in her seventh decade. Her choice of words revealed her struggle to understand the disease and to be sure it was not cancer. As others did, she took comfort in deciding that the epileptogenic brain area is small:

> It's the vocabulary [that's difficult to understand], and that's part of me seeing the picture of where it [the epileptogenic tissue] is and what it is. It's not a huge thing. It's just one little, teeny, tiny block.

A high-functioning woman described many inner conversations which included views of herself as an efficient person that conflicted with personal regret about her years of ignorance and denial concerning the cause and course of her epilepsy. If life could be lived again, participants reflected that they would be more proactive in seeking treatment and not simply accept or "forget" about the epilepsy.

Receiving care at a specialized epilepsy center stood in sharp contrast to past treatment experiences for the majority of our participants. Perceptions of severity were framed around care that previously lacked direction. Once at an epilepsy center, seizures that changed the state of the brain and diminished personhood were finally taken seriously by a team of clinical experts. In a few simple words one person said that working with a specialized team gave her "a sense of going forward [with her care] versus a sense of a wall."

Participants described their epilepsy as a complex disorder of a master organ that was hard to diagnose and hard to treat. Given the added concern that ongoing seizures increased functional risks to the brain the option of epilepsy surgery was considered. For many participants the need for brain surgery underscored the severity of the illness. For the subgroup of participants who were awaiting surgery, signing the informed consent represented an optimistic turning-point. However, many participants also reflected contradictory views of illness severity as they weighed the option of surgery. Personal judgments that seizures were not bad enough to warrant brain surgery created hesitancy around surgical options. On the other hand,

participants also reasoned that if the epilepsy warranted brain surgery, then the seizures could no longer be dismissed, and the severity of illness had to be admitted. An inner tug-of-war captured what our participants wrestled with as they defined their personal values and identified what was important to them as they made treatment decisions.

A long and "uphill" course of illness represented a difficult personal struggle for all participants. They concluded that personal maturity was needed to accept a condition that was expected to be lifelong. Accepting the epilepsy was considered important to personal happiness and integral to healthy adjustment. But, in this process of acceptance, participants downplayed the seriousness of illness and often pushed the illness into the back of their minds.

Resisting Severity

Seizures, the pivotal experience of epilepsy, were often hidden from the self because participants were not conscious during seizures. Many relied upon witnessed accounts, or rationalized that a seizure had occurred because of gaps in time, or postictal symptoms. Very few participants had seen their own seizures and only four knew others diagnosed with a seizure disorder. Not wishing to exaggerate their own epilepsy, most thought that other people had comparatively worse seizures than they did. This contributed to participants resisting the realization of how severe their seizures actually were. An example was provided that illustrated how a young woman planning a graduate school career rationalized the severity of her epilepsy:

> Seeing that someone has something worse than you, it makes you kind of reevaluate. It made me kind of reevaluate my own sadness and unsettledness about my, um, my situation. It was like, you know, it's not that bad….I've never been suicidal…It's just a deep sadness inside that is sometimes hard to get rid of.

Furthermore, participants compared DRE with other chronic illnesses such as cancer or childhood disabilities and determined that their epilepsy was not the worst condition possible. Many positive gains were attributed to having epilepsy. These gains included learning patience and empathy, overcoming shyness and becoming a generally nicer person. Pride was also

taken in the personal strength that was required to manage a complex illness. In the context of life-long epilepsy, the motivation to cure the epilepsy was dampened by the uncertainty of trading a familiar life for an unknown future without seizures. One person with seizures since childhood viewed his epilepsy as, “[Epilepsy is] who I am. So, it comes in the whole package. It's one more thing that you have to deal with”. However, when epilepsy was accepted as a condition of life, a sense of urgency was less apparent, and severity was downplayed in deference to coping well.

Rating illness severity. A direct question about subjective ratings of illness severity on a 10-point scale was difficult to answer as participants vacillated about their ratings and resorted to a relativist approach. Considering how they would rate the severity of their epilepsy, decisions and actions taken in the past were evidence of thinking about and interpreting their personal situations. In general, the epilepsy was rated as less severe when seizures were not outwardly noticeable or when they did not occur every day. Epilepsy associated with injuries and the need to wear a helmet was considered to be more severe. In addition, the level of oversight required by a family member also factored into severity ratings.

Resisting limitations, and not giving in or waving the white flag played a role in how severity was regarded. Some reasoned that it was safe to drive if seizures were so subtle that others did not notice them, or if the aura was long enough to allow the driver to pull over before losing awareness.

Another illustration of the challenge of subjective ratings was that chronic medical conditions overall were identified as comparatively worse than epilepsy including asthma, kidney disease, multiple sclerosis, cancer, and not being able to walk or talk. A participant who exemplified this finding described his lifelong seizures as “worser than diabetes…and comes close to a heart attack”, yet he rated his illness severity as 5/10. This rating was surprisingly low considering that he suffered from clustered seizures, required frequent emergency room visits and was at great risk of falling.

The possibility of dying from seizures was frequently minimized by participants along with downplaying the urgency to improve seizure control. Perceptions about dying from seizures varied from the idea that death was possible but unlikely, to statements of fact such as, “I have not died yet”. One participant regretted not asking her doctor, “Can I die? Because these seizures are getting worse. Can I die?” Not waking up after a nocturnal seizure was considered to be a real possibility. While several participants knew about that sudden unexpected death in epilepsy (SUDEP) is a possible consequence of uncontrolled seizures, most did not feel that they were personally at risk. To illustrate the subjective context that represented the risks of epilepsy, a man with seizures since early childhood, rated his epilepsy severity as 9/10 because he had required treatment in the intensive care unit for seizures in the past and someone in the family had succumbed to SUDEP. However, despite a high severity rating, he reasoned that his life was “pretty normal”. He did not associate seizure risks with disability, but defined himself as “just your everyday person.”

Overall, attempts to rate disease severity on a 10-point scale led our participants to describe that the impact of seizures on daily life was all-encompassing. Various factors were taken into account when participants attempted to determine severity. Unpredictable fluctuations in frequency led many to be indecisive about a selecting a specific number on a rating scale. This indecisiveness was illustrated by a young mother who initially rated the epilepsy as low (4/10) because her life was plentiful, and she did not feel limited. However, she vacillated and changed the rating to 8/10 because of the frequency with which she lost consciousness and her worries about the future. This participant, who had lived with seizures for more than two decades, illustrated the internal battle in determining severity when she said:

> There is no cure. You’re going to have it for the rest of your life, so you just need to come to understand that you have to deal with it. And there’s people worse off that have certain things… So, try to be happy that you don’t have some of the conditions that everyone else has.

On the other hand, another participant who had seizures for more than four decades was decisive about his 10/10 rating. He reflected upon his thought processes by saying:

> It's a ten because …it is definitely impacting my life on many levels. So, for me, it is a huge impact. Now if you ask me, am I dealing with it? Yes, I am. Is it really impacting to the level I cannot live? No, I think I am managing pretty well. Does it bug the hell out of me? Yes, it does.

Experiencing Burden: "A Burden on My Back"

The weight of living with an illness that was unseen to the self yet created restrictions and dependency was described as burdensome. One participant said:

> I realize I'm missing a huge chunk of human experience and probably good things in my life. And when I get to this age and look back and see all the things I've missed because of the fact that I lived with this all my life ... Life, like everybody else's but for me especially, was a struggle getting to where I am because there is this huge, you know, burden on my back.

Burden was an umbrella term that encompassed mutual suffering experienced by participants and their families. Speaking collectively one participant said, "We are tired, and weak and drained…we are fighting with ourselves every day." The properties of burden took many forms framed around the impact of illness on the self, close family and society.

Learning to live around DRE and coming to the realization that cure was elusive occurred over time. A partial response to medicines i.e. fewer seizures had been accepted by physicians and participants prior to arriving at a specialized center and was integral to a disorder in which uncertainty and "just not knowing" were expected components of the clinical trajectory. The chances of being a burden on society were reduced when it was considered that a reasonably "normal" life was being lived. While participants initially resisted the impact of uncontrolled seizures, they eventually gave-in, "waved the white flag" and accepted the epilepsy. Acceptance served to validate personal life-circumstances. Although some did not

identify with the sick role, there was practical value to accepting the sick role as a way to lessen burdensomeness and limit embarrassment.

With respect to the family, an aspect of hardship was centered on shifting family roles. A wide circle of family relationships was affected as an increased burden of responsibility was placed on spouses, parents and children. Parents were especially conscious that children might experience distress when witnessing their seizures and post ictal confusion. Most participants were aware of the care they needed at home, but the necessity of vigilant oversight created a mutual burden. Sometimes repercussions of family dissent led to feelings of encumbrance. Speaking about her grandmother, a young participant said:

> She starts criticizing my mom, but she doesn't see how much she takes care of me or what she goes through, seeing me [when seizures occur]. And that kind of gets you mad and sad at the same time, you know. So, there are times that you can feel like you're such a big deal, you know, or like such a burden.

While the experience of burden was not specifically asked about during the interviews, burdensomeness was openly and deeply described. The degree of burdensomeness was an unexpected thread that stretched across all four categories of our data. Participants had grown tired of depending on others and travelling as passive members of their households. High dependency left many participants feeling marginalized and defenseless. Much burden was created around social limitations related to not driving, the need for oversight and the constant need to monitor personal well-being. Many anticipated that surgery might restore the freedom of spontaneous living that had been lost. Ultimately, burden was equated with the experience of deep sadness. Treatment delays and missed opportunities brought disapointent, families were discouraged, and social isolation created unhappiness. Therefore, sadness contributed to evidence of burden due to loss of autonomy, a sense of inertia and resignation around the need to live with and tolerate ongoing seizures.

Key Points

The notion of illness severity in our sample was not uppermost in participant's minds. On the contrary, perceptions of severity were arrived at after much discussion around the impact of illness on daily life. An internal struggle existed between the opposing forces of admitting and resisting that a serious condition existed, and balancing the burden. In the past, seizures had been ignored or minimized and participants rationalized that other people with epilepsy were worse off. Furthermore, other medical conditions were considered comparatively worse than epilepsy. When asked to expand on personal responses, the physical, social and emotional impacts of uncontrolled seizures for the self and the family were perceived as a burden that could no longer be tolerated. Participants reasoned that worsening seizures carried risks to the brain and that life was on hold, therefore the seizures had to be more urgently addressed.

Conclusion

The representations of illness severity in participants with diverse clinical and sociodemographic characteristics who had lived with DRE for an average of 15.4 years were highlighted in this chapter. Many facets of the broader illness experience were illuminated that ultimately framed perceptions of severity and gave voice to the thoughts and actions of our sample. The impact of unpredictable seizures on daily life was substantial, but participants did not naturally think about the severity of their epilepsy or consider it as serious, severe or urgent.

Four dynamic and interactive categories encapsulated perceptions of illness severity and how this was arrived at in the minds of participants. Tensions, that is contradictions, indecisions and ambiguities, were revealed within and across the categories to reflect whether and how participants described severity. A sense of illness severity was arrived at through a process of gradually unpacking these multifaceted experiences. However, when participants took a comparative view of their epilepsy, the severity of personal illness was underrated.

The experience of “epilepsy is an uphill thing” represented the first category. Life had to be lived despite ongoing seizures and around many limitations. Epilepsy was perceived as a lifelong condition, yet the restrictions it imposed meant the future was unclear. It was concluded that treatment lacked direction and life was on hold.

The second category, “It’s just not knowing”, reflected the uncertainty of living with a condition characterized by unpredictable lapses of awareness. It was difficult to understand the symptoms of a condition that were often hidden to the self, yet created disability that was often not outwardly appreciated by others. Pride was taken in developing self-management strategies, however, normalizing and minimizing the illness may have served to reinforce passive acceptance. Disclosure was a powerful component of normalizing the epilepsy. Some participants viewed seizures as major events that should be disclosed, but for others efforts were taken to conceal the condition and under report the seizures. To avoid injury and embarrassment, precautions had to be taken that meant life was lived with caution.

The third category, “waving the white flag”, captured the essence of having to depend on others for safety and survival. For our sample, episodes of loss of awareness meant that bodies and minds could not be relied upon. Losing autonomy required submitting to others for help in many areas of life, especially when seizures occurred. Portrayed as being “at the mercy of others” there was little choice but to concede that vigilant oversight was necessary. Giving over control of key aspects of living to family members created mutual suffering.

At this point in the process a sense of urgency was revealed. The fourth category, “battling with myself”, reflected the mutual burden experienced around a condition that created a diminished self, and took a toll on the family. Up until now many efforts had been taken not be overwhelmed by the epilepsy or to be defined by it. However, much internal debate centered on admitting notions of severity yet resisting it at the same time. Participants avoided notions of self-pity and downplayed the risks of dying from seizures. Most decided that other medical

conditions were worse than epilepsy, and that other people had worse seizures than they did. This relativist perspective kept perceptions of severity contained.

Chapter 6: Discussion

The illness experiences of participants with focal drug-resistant epilepsy (DRE) who presented for treatment at a specialized, academic epilepsy center were explored with constructivist grounded theory (CGT). The specific focus of the study was to understand how participants viewed their illness and the ways in which perceptions of illness severity were represented. The data was comprised of 51 interviews in 35 adults, of whom 20 (57%) were women. Our sample included participants with diverse clinical and socio-demographic characteristics, who had lived with uncontrolled seizures for an average of 15.4 years (range 2-44 years).

The data collection and analysis stayed faithful to the systematic guidelines of CGT methodology and included semi-structured interviews that were coded in multiple steps. The assumptions that underlie this study were based on the backgrounds and professional experiences of our research team, specifically my many years as an advanced practice nurse providing care to patients with DRE and their family members. These assumptions included that 1) drug-resistant epilepsy (DRE) is a serious, under-recognized disorder, 2) early recognition of DRE and timely referral to a specialized epilepsy center may have the potential to disrupt the trajectory of illness experience, and 3) multiple views exist around individual illness perspectives that shape the decisions people with epilepsy make and the actions they take.

Illness severity was implicit in our data and was gradually realized as participants reflected upon the meaning of their experiences at four levels that were interrelated and co-constituitive. Core components of this process are presented in a theoretical framework (Appendix G) with four categories and subcategories. Collective experiences around uncertainty, shared suffering and burdensomeness were situated in the core categories of “Epilepsy is an uphill thing”, “It’s just not knowing”, “Waving the white flag” and “Battling with myself”. An evolving process involved appreciating the burden for self and others. It culminated in the conclusion that an urgent need existed to better address the epilepsy.

“Epilepsy is an uphill thing”: While DRE is recognized as serious among clinicians, our participants appeared reluctant to describe their uncontrolled epilepsy as severe. Part of the “uphill battle” of living with DRE was coming to terms with an illness that was poorly understood and not familiar to the general public. A long course of drug resistance, perceptions of unlikely cures and the restrictions imposed on their lives left participants feeling trapped and “stuck” participants

Reflections about encounters with the medical system were fraught with criticisms related to rushed and impersonal clinic visits that were focused only on seizure frequency and medicines. These common experiences led to a sense of treatment inertia and disempowerment similarly documented in other studies (Dewar & Pieters, 2015; Keikelame & Swartz, 2016; Varley, Delanty, Normand, & Fitzsimons, 2011). Added to this, perceptions of low disease severity may have encouraged behaviors such as hiding the epilepsy, deliberately under-reporting the frequency of seizures, and continuing to drive against medical advice.

While participants desired more definitive treatment most did not know that epilepsy specialists existed and some accessed our specialized epilepsy center by chance. While obtaining care at a tertiary epilepsy center was seen as an opportunity to consult with clinical experts, the meaning of obtaining medical care at such a center was not linked to the severity of the disease. Notably, the desire for more aggressive treatment was also not highlighted by patients, care givers or clinicians in a study that explored treatment priorities in epilepsy (Thomas, Hammond, Bodger, Rees, & Smith, 2010). While a lack of urgency may reflect the practical implications of treatment uncertainties intrinsic to the management of chronic epilepsy, this attitude also seems to underscore an overall sense of treatment inertia in epilepsy care.

Surgical candidacy for our participants was an important milestone in a complex and long journey. However, while many patients do embrace the option of surgery, studies report that 21-28% of surgical candidates declined surgery although the reasons for this remain unclear studies report that 21-28% of surgical candidates declined surgery although the reasons

for this remain unclear (Anderson, Noble, Mani, Lawler, & Pollard, 2013). We suggest that the willingness to embrace surgical treatment may be tied to the social meaning of health, illness and disability and individual interpretations of living with chronic epilepsy. Across examples of chronic illness experiences, personal views that conflict with treatment recommendations reflect important insights into how the intrusiveness of chronic illness is managed (Charmaz, Harris, & Irvine, 2019). Perceptions of illness severity may be foundational to decision-making yet ways to evaluate these perceptions in a clinical setting have not been explored.

"It's just not knowing": The second category, "It's just not knowing", encapsulated the need to live despite active epilepsy and reflected the uncertainty of life with chronic seizures. Living around unpredictable seizures was about developing strategies for self-management. Adaptive strategies included normalizing and minimizing symptoms, but also required taking precautions and living with caution. Participants spoke of "taking epilepsy in stride" or "forgetting about the epilepsy", yet life with seizures was perceived as stressful. The need to take safety precautions and always plan ahead meant a normal life could not be lived and daily life lacked spontaneity. At the center of a personal dilemma was the issue of whether to hide or disclose seizures.

Passive acceptance was evoked by long-standing epilepsy and observed in our participants when it was perceived that nothing more could be done. As shown in other studies, perceptions of limited control negatively impacted the course of illness and call for improvements in the delivery of care to encompass what patients believed about their disease (Elliott, Lach, & Smith, 2005; Ji et al., 2016; Varley et al., 2011). Some participants did not think their epilepsy could be cured because the brain was assumed to be too complex. The only remaining alternative was to "manage" the epilepsy and live around it, or in spite of it. As found in another grounded theory study (Schneider & Conrad, 1983), our participants struggled with disease limitations but worked hard to find ways to adapt and normalize life. Hiding and minimizing seizures was observed across the participants as a strategy commonly used to cope with stigmatizing symptoms in many chronic illness (Charmaz et al., 2019).

Although intermittent seizures were a constant reminder of the epilepsy, severity of illness did not appear to be uppermost in the minds of our participants. The risk of dying from seizures was minimized and was not associated with personal risk. Our findings were highlighted in a novel qualitative study that explored how meaning was attached to understandings about the risk of sudden death in young adults with epilepsy (Harden, Tonberg, Chin, McLellan, & Duncan, 2015). On a superficial level tolerance of seizures and acceptance of epilepsy were viewed as components of coping well. Revealed as a tension in the analysis participants simultaneously expressed concern about worsening seizures, contralateral spread in the brain and cognitive decline. Ultimately, participants reasoned that the epilepsy could no longer be ignored.

"Waving the white flag": Notions of severity accumulated around the social impact of an illness that had been long fought. Diminished autonomy linked with the loss of self-reliance meant depending on the support of others, as represented in the words "waving the white flag". Reliance on others evoked existential ambiguity as illustrated by one person who said, "You're being told how to live a life that's not yours to live." Participants agreed that assistance with tasks of daily life and constant surveillance were necessary to safety and survival. However, submitting to the care of others meant living at the pace of others and came at the price of becoming socially isolated. The impact of dependency on participants and family members in our study was seen as part of a sad and lonely struggle that created mutual suffering. These distressing representations were highlighted in the findings of a qualitative study (Yennadiou & Wolverson, 2017), and were also noted as a component of burden of care for family members in a recent review paper (Saada, Wang, & Bautista, 2015).

Participants were motivated to achieve seizure control as a way to overcome mutual suffering. Although brain surgery was a frightening option, many participants were willing to sign a surgical consent for the sake of their families, even when some family members were very hesitant and actively discouraged surgery. Many conflicts were apparent in our data around how

families viewed the seriousness of the epilepsy and how they influenced processes of treatment decision-making.

“Battling with myself”: The intermittent nature of seizures and symptoms that were often not apparent to the participant meant it was possible to ignore the epilepsy. A “battle with myself” captured the essence of the internal conflict that occurred around weighing the severity illness and ultimately what to do about it. Three elements were at the center of this battle: admitting severity, resisting severity and experiencing burden. Admitting illness severity took into account composite aspects of experience, such as the amount of time lost to seizures, perceived risks to the brain, the need to take anti-seizure drugs that seemed futile and the restrictions imposed on everyday life. In contrast, participants actively resisted thoughts of illness severity that equated to self-pity. Pride was taken in living around the limitations of disease.

A notable finding in our data were the many references to sadness. The condition of epilepsy was openly and spontaneously referred to as “sad” and even tragic. Sadness was reported around the ineffectiveness of anti-seizure drugs, the embarrassment of seizures and feeling abandoned by a medical system that appeared to accept uncontrolled seizures as inevitable. Deep unhappiness extended to the impact of epilepsy on significant others as a marker of disease burden. It was also an emotion that motivated efforts towards improved seizure control and improvements in general health. The word “sad” was used to reflect personal suffering and to highlight a human response to the illness experience. During the interviews, participants were given the opportunity to step outside of the narrow clinical focus, that of seizure frequency and anti-seizure medicines, to reflect on how life was lived with the condition itself. This observation illustrated the power of qualitative research to understand the personal meaning of illness experiences.

Participants were despondent about unmet developmental milestones that left their lives restricted and the future uncertain. A sense of personal tragedy expressed by our sample was

also highlighted as part of the experience of DRE in a study of the illness experiences of adolescents (Elliott et al., 2005).

Three aspects of burdensomeness stood out as a key component of perceived severity. Firstly, opportunities for reciprocal living were reduced. Secondly, developmental goals were disrupted, and participants determined they had to work twice as hard to achieve their dreams. Thirdly, epilepsy was an obstacle that limited normal life and presented unique challenges for the self and others. Irrespective of age, the risk of untimely seizures was encountered as a social burden that cost friendships and job opportunities. Our participants rejected all notions of self-pity, but acknowledged the worry, fear and helplessness experienced by those around them.

Our participants expressed difficulty rating their personal severity of illness and spontaneously searched for comparisons with other disease conditions or what they imagined were more serious seizure types. Other people's epilepsy was often perceived to be worse and the frequency of convulsive seizures as opposed to subtle focal seizures also made a difference. By taking a relativist view our participants lessened their personal urgency. Severity ratings were often surprisingly low considering the risks of life-threatening complications including status epilepticus, and sudden death. Overall, participants worked hard to pursue identities that avoided disability, and to minimize severity. Despite the challenges, epilepsy was not always regarded as a negative life experience. These gains that included personal resilience and compassion enriched personal biography, a phenomenon described in other epilepsy studies (Luyckx et al., 2018; Schneider & Conrad, 1983) and as a general feature of chronic illness experience (Charmaz et al., 2019).

Intrinsic Tensions

The dynamic tensions that were evident between categories and within subcategories were both surprising and contradictory. Six intrinsic tensions are individually discussed namely, 1) epilepsy was a condition that was unwanted yet had to be owned, 2) despite the

extraordinary challenges of DRE, participants viewed themselves as ordinary people, 3) DRE represented many struggles, yet the condition was not directly referred to as severe, 4) younger versus older age of onset may influence perceptions of severity, 5) risks of dying from seizures were downplayed and 6) adapting to chronic epilepsy may actually be maladaptive in the long run.

Participants in the present study related unwanted feelings of loss of dignity and reduced self-worth to not being able to trust the brain. The impact of a sense of body failure in epilepsy has been associated with self-management strategies around coping with a threat to self-concept in previous research (Kilinc, Campbell, Guy, & van Wersch, 2018). To overcome feelings of defeat and to achieve personal goals, sustained efforts were required to “fight the epilepsy”. In this process, the epilepsy appeared to be positively assimilated into the lives of our participants. Despite extraordinary challenges, many participants referred to themselves as ordinary people who lived normal lives and did not want to be seen as disabled, or in need of help. Striving to normalize the epilepsy required talking about it, and not hiding or ignoring the condition, although much conflict around disclosure is documented in other qualitative studies (Elliott et al., 2005; Rawlings, Brown, Stone, & Reuber, 2017).

Many distressing aspects of epilepsy were experienced, but DRE was not directly referred to as severe. Life had to be lived and ways had to be found to overcome limitations that were captured in phrases such as fighting harder than others in order to succeed. Attitudes and beliefs played a role in how the illness impacted self-concept to influence the ways severity was represented in our study. Linked to our finding, a series of spontaneous, written accounts by people with epilepsy did not include perceptions of urgency or severity, leading the authors to believe that people did not divulge the true size of the problem they lived with (Rawlings et al., 2017).

The purpose of our study was not to compare subgroups however, epilepsy that began at different developmental times appeared to have varied implications for illness acceptance

and impacted life in both similar and different ways. Among those whose epilepsy began in adulthood, restoring life to previous levels of independence was an overriding goal. For those with epilepsy since childhood the goals were to establish independence and to live a “normal” life. Where younger subjects with epilepsy reported greater anxiety and higher disability scores than older subjects in one study (Sajobi et al., 2015a), older subjects in another study experienced great distress due to added limitations of advancing age (Yennadiou & Wolverson, 2017). The impact of body failure for self-concept may be greater when epilepsy begins in adulthood (Kilinc et al., 2018). This comparative observation imposes a new layer to understanding how the severity of illness is expressed that deserves to be more deeply explored.

The possibility of dying due to seizures was downplayed and the risk of SUDEP was not prioritized as a reason to seek improved treatment. Furthermore, the risks of brain surgery were often thought to be greater than the risks of ongoing seizures leaving some people to be hesitant to go through with it (Choi et al., 2011; Erba, Moja, Beghi, Messina, & Pupillo, 2012). Interestingly, while the bothersomeness and uncertainty of seizures were included as items in the recently developed Personal Impact of Epilepsy Scale (PIES), worry about dying from seizures was omitted (Fisher, Nune, Roberts, & Cramer, 2015). It was suggested by previous researchers that unexpected death represents yet another area of life over which patients have no control leaving many to separate the possibility of SUDEP from the wider meaning of epilepsy experience (Harden et al., 2015).

The last tension, taking pride in adapting to life with seizures, may be maladaptive in the long run. It was striking as reflections that life had been well managed despite the epilepsy, reduced the incentive to view the epilepsy as severe. Using grounded theory methodology as in the present study, a previous researcher reported that participants who “manage[d] their epilepsy to greatest advantage were those who defined it as of minimal importance to them” (Schneider & Conrad, 1983, p. 232). Since, minimizing symptoms also had implications for

disease outcome in the present study we believe this should be directly addressed as an important issue when counselling patients about disease management.

Components of Illness Severity

We extrapolated that severity was framed around four domains that represented the impact of illness on physical, cognitive, social and emotional aspects of life. Properties of each domain may serve as a framework for the future development of an instrument to measure perceptions of severity based on the descriptions of participants living with DRE.

The first domain relates to the physical impact of seizures. This includes many elements of seizure severity reflected in standard measurement tools such as ineffective drug therapy, the fear and worry of unpredictable seizures, the duration of seizures, time of day they occur and the type of seizure.

The second domain involves the cognitive impact of illness. Elements of this domain include the confusing aspects of remissions and relapses, moodiness, anxiety, cognitive decline and memory loss. The experience of becoming resistant to medicines meant that cure was not possible. At the same time, side-effects of anti-seizure drugs were experienced as worse than the seizures.

The social domain encapsulates burdensomeness. This domain includes mutual suffering experienced around seizure events that were experienced as reportedly traumatic for everyone. Not being able to rely upon oneself for daily functions meant becoming dependent on others. A loss of personal autonomy was coupled with constantly waiting for assistance and becoming socially disconnected.

The fourth domain includes the emotional elements of running out of treatment options, experiencing sadness and determining that daily life has lost a sense of spontaneity. The final elements of the emotional domain include that life is restricted and the future is on hold.

Conclusions and recommendations

Illness severity in DRE is a concept that goes beyond episodic seizures to include the social and psychological impact of a multifaceted experience. At present existing markers of severity include clinical markers such as seizure frequency and number of anti-seizure drugs. Illness severity as a marker of overall disease experience goes further than clinical markers yet has only recently received attention (Sajobi et al., 2015b). Based on the findings of the current study, how to measure the multiple domains of perceptions of severity of DRE represents a new challenge for outcomes research. Another marker, that of burdensomeness appeared to be interwoven throughout our analysis. The burden of illness as a marker of severity may serve to encompass the dimensions of uncertainty and dependency evident across the categories.

Contradictions, conflicts and uncertainties were revealed across the categories and between the subcategories. Participants spoke about treatment that had been directionless and that their lives that were on hold, however once aware of other options, the timing of treatment decisions was one area of life in which participants retained a measure of control. It is also possible that self-reports of severity may not be a definite tipping point when it comes to following through with proposed treatment as demonstrated by participants who signed a surgical consent but later chose not to undergo surgery. As such, the power of illness perceptions to impact the trajectory of illness was highlighted.

A conceptual model that was based on a review of qualitative literature captured a broad network of interconnected areas in which epilepsy impacts life (Kerr, Nixon, & Angalakuditi, 2011) without consideration of severity. The present study showed how a complex set of experiences come together for people living with DRE that has relevance for what individuals say they do about it, and when and how treatment decisions are made. Perceptions of severity may take a network of illness impacts one step further to create overall meaning that motivates treatment decisions. Therefore, perceived severity may ultimately impact the urgency with which

treatment decisions are made and the timing of such decisions to reveal ‘severity’ as an endpoint in itself.

Although results of a mediation analysis showed that seizure freedom, depression and anxiety were determinants of epilepsy severity (Sajobi et al., 2015b), single global measures of epilepsy severity (the GASE scale) and perceived disability (the GAD scale) may not take into account the true impact of epilepsy (Raty, Soderfeldt, Larsson, & Larsson, 2004). In our study, eliciting a score on a 10-point scale that reflected severity was not easy for our participants. Based on this experience we argue that illness severity is a construct that cannot be meaningfully established using a single, global score. To get at the core subjective meaning of symptoms and their impact on social interaction required an inductive process that arose from carefully teasing apart the domains of personal experience highlighted above. Our findings based on a substantial data set with rigorous data analysis provides the foundation for the development of a new tool to measure severity that can be used in a busy clinical settings and as a research tool.

Illness identity lay at the heart of how illness was represented and appeared to be an important factor that motivated a sense of treatment urgency. How patients identify with their epilepsy may facilitate, delay or even negate treatment decisions and has been recommended as a target for individualized treatment interventions by previous researchers (Luyckx et al., 2018). By means of abductive processes we have gone beyond a description of illness experience. We explored subjective perceptions of severity as a way to better understand the course of illness experience in our participants and to identify domains central to a process of recognizing the severity of illness that evolved over time. Because the psychosocial challenges of DRE are more burdensome than the clinical factors (Borghs, Tomaszewski, Halling, & de la Loge, 2016; Shallcross et al., 2015) it is time to move away from a disease-oriented model towards a more holistic model of care (Rawlings et al., 2017). To reduce the burden of a serious

illness and promote timely access to specialized care, an important opportunity is presented to integrate the biomedical and the social sciences.

Study Strengths and Limitations

Our study afforded foundational insights into an important yet under-researched phenomenon in epilepsy. The richness of our findings were a function of conducting semi-structured interviews rather than a structured questionnaire and served to highlight the strengths of qualitative methodology for research concerning human experiences of chronic illness (Morse, 2016). A strength of this research includes the diverse socio-demographic characteristics of the sample (Corbin & Strauss, 2015, p. 141). Another strength is that the 35 participants are larger than common estimates of between 20-30 participants (Mason, 2010, p. 3). Analytic rigor was enhanced and bias reduced as the interviews were conducted by the same interviewer (Dr Huibrie Pieters), a skilled qualitative interviewer, who was purposefully selected as she was not part of the clinical epilepsy team (Hesse-Biber, 2017, p. 349). The data analysis that included systematic memo writing, coding and categorizing was undertaken by two researchers. Constant comparisons and discussions between researchers confirmed interpretations that facilitated the dependability and credibility of the findings (Houghton, Casey, Shaw, & Murphey, 2013). Another important strength of the study is that the sample was clinically heterogenous, and included a range of verbal comprehension scores that enabled us to hear the voices of participants with varied intellectual abilities. While participants represented a range of socio-demographic characteristics, a limitation of the findings is the small percentage of participants who self-identified as Asian or African American. Another limitation is that recruitment was limited to a purposive sample of patients with focal DRE undergoing treatment at a level four, epilepsy center in an academic medical center located in a large metropolitan US city. The resources available at other urban or rural settings versus those at a major academic

center may shape illness perspectives and effect decision-making in different ways. Presenting at a specialized center for treatment may in itself influence patient perceptions of illness.

Clinical Implications

The research was based on the assumption that patients' perceptions of illness severity have vital implications for outcome patterns in uncontrolled focal epilepsy. This qualitative study was undertaken to fill a gap in understanding that may explain how patients tolerate chronic seizures for upwards of two decades before presenting at specialist epilepsy centers. A finding with overarching clinical implications is that illness severity, while discussed among epilepsy specialists, did not appear uppermost in the minds of our participants. Although seizures were described as severe, determining the severity of illness was much more complex and was tied to the social impact of disease. It was only when awareness of the functional and practical impact of uncontrolled seizures on daily life were raised that severity was acceded to. Several implications are deduced from the findings that pave the way for improvements in clinical practice, nursing science and public policy.

Perceptions of illness severity in our sample were impacted by the messages participants reflected hearing during clinician-patient interactions. With respect to clinical practice, while the course of epilepsy is hard to predict, diagnostic and treatment language should clearly articulate the seriousness of DRE as symptom severity and treatment urgency is negatively impacted in the absence of such direct communications. The risk of sudden death, an inherent risk of DRE was rarely referred to in this study. Thus, open clinical conversations about the seriousness or DRE and the risk of death will likely also promote patient's perceptions of severity.

An all-encompassing clinical implication of the study is that the perspectives of both patients and clinicians with respect to treatment urgency played a vital role in shaping the illness trajectory. Since personal narratives have much to teach about the intersection between how

disease is understood and how illness is experienced, it is important to find ways to illicit the patients' viewpoint. The language of epilepsy was difficult for participants to understand, to process and to remember. Even though participants resisted dependency, family members share the burden of illness and were relied upon to interpret clinical encounters and remember the plan of care. Resources such as MRI or video evidence to support explanations need to be used with care as it is possible for people to misinterpret professional explanations. Not having seen video recordings of their seizures made it possible to ignore the disorder, although many participants stated no desire to see their seizures. For those who had seen their brain MRI, determining that the area of brain tissue responsible for seizures was small, contributed to minimizing the epilepsy. On the other hand, this visual evidence encouraged some participants to go ahead with surgery.

Epilepsy self-management is generally focused on practical aspects of epilepsy care such as seizure and lifestyle management and adherence to medicine prescriptions, but much less emphasis is placed on building active partnerships between clinicians, patients and families or carers. Patients and families require guidance to understand a confusing illness and navigate a complex health care system in order to benefit from timely treatment. For this reason, it is necessary to include family members as part of the treatment team.

Promoting awareness of illness severity in the minds of patients and clinicians has implications for the establishment of priorities in epilepsy care and the direction in which treatment is steered. Clinical partnerships need to be fostered to promote shared decision-making and reduce unnecessary chronicity. It is important that clinicians are sensitized to the negative consequences of minimizing the severity of DRE and a tendency to justify passive acceptance. Our findings highlight the responsibility of multidisciplinary leadership in epilepsy to challenge attitudes of acceptance around uncontrolled seizures and to communicate standards of care that reflect timely interventions.

Nurses specialized in epilepsy are an underrepresented group within this neuroscience subspecialty. The professional contribution of advanced practice nursing in neurology and neurosurgery is gaining traction across the country as evidenced by increasing numbers of nurse practitioners staffing community-based neurology clinics. Since formal training for nurses in the neurosciences and in epilepsy is limited, the communication of knowledge gained from the findings of this research is crucial for shaping attitudes and standards of practice.

With respect to public policy, efforts are needed to increase professional and public awareness of the scientific advances in epilepsy and the expanded therapeutic options. Urgent attention needs to be given to solving medical system failures that contribute to disability, namely, the slowness of referrals for specialty care. The lack of public awareness of epilepsy stigmatizes and marginalizes people that live with seizures, reinforces a hidden disorder and contributes to the severity of illness.

Several implications for future research have been identified. The purpose of the present study was not to compare groups. However, as there were indications that differences in perceptions of severity were influenced by age at onset this is an area for future research. In addition, despite acknowledging that illness is severe, patients often delay or decline epilepsy surgery once eligible. The reasons for this unclear and deserve to be explored. A better understanding of decisional processes that identifies ways to recognize social and cultural hesitation early in the surgical process may help to guide appropriate counselling about key areas of epilepsy care.

Instruments that measure illness severity are crucial for epidemiological studies, clinical trials and practice (Thurman et al., 2011). Because it can be difficult to establish individual levels of disability, practical and validated tools to guide the assessment of illness severity need to be further developed. Our qualitative findings lend credence to components of severity that might be tested to develop a practical tool that is sensitive to severity. Finally, the importance of family care givers is highlighted as a core component of illness experience, yet the challenges of care

giving, and the support needed for care givers is an under-studied area in epilepsy and another important area of potential research.

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Appendix A: List of Abbreviations

AEDs	Anti-epilepsy drugs
CDC	Center for Disease Control and Prevention
CGT	Constructivist grounded theory
DRE	Drug-resistant epilepsy
EFA	Epilepsy Foundation of America
GT	Grounded theory
ILAE	International League Against Epilepsy
NAEC	National Association of Epilepsy Centers
QoL	Quality of life
SI	Symbolic interactionism
SSIG	Semi-structured interview guide
SUDEP	Sudden unexpected death in epilepsy

Appendix B: Summary of Clinical and Social Trajectories in Epilepsy

Article	Year	Summary
Sajobi et al.	2017	Purpose: Traced the course of new-onset epilepsy in children across one of three distinct trajectories based on quality of life (QoL) scores in four domains. Findings: Baseline behavior and cognitive problems in children and depression in parents predicted poor QoL outcomes in children 24 months after diagnosis.
Choi et al.	2016	Purpose: Utilized retrospective chart note reviews to classify trajectories of seizure fluctuations into four patterns in adults who failed a second anti-seizure drug. Findings: Focal epilepsy (72%) and encephalopathic generalized epilepsy (93%) were the most refractory. Longer follow-up time was associated with better trajectory patterns.
Ramsey et al.	2016	Purpose: Identified 'at risk' QoL domains in new onset epilepsy in children measured over 24 months in order to establish a set of most critical predictor variables for QoL. Findings: Four trajectories were identified. The number of anti-seizure drugs was the most consistent predictor of poor QoL, along with depression and adherence. Interventions that improve family function were recommended together with cognitive-behavioral therapy.
Pieters et al.	2016	Purpose: Described the "arduous" experience of time before referral to a comprehensive epilepsy center among parents of pediatric patients. Findings: Four themes characterized parental experience. Modifiable system barriers may improve provider-based and parent-based decision-making that will ease a difficult treatment journey.
Baca et al.	2015	Purpose: Explored the time course to surgery and the barriers to surgery in post-surgical pediatric patients as perceived by parents. Findings: Epilepsy surgery was ultimately perceived as a hopeful and necessary option. Navigating system barriers contributed to treatment delays. Influence of parental attitudes to timing of surgical referrals and undergoing surgery needs to be explored.
Berg et al.	2015	Purpose: Determined the patterns and frequency of remissions and relapses over a 20-year period in 619 newly diagnosed children. Findings: Patterns of remission and relapse were highly variable and difficult to summarize. Over 20 years, >50% achieved complete remission, but the course was very variable and depended on type of epilepsy. Of those who failed a second anti-seizure drug, 25% did so four years after diagnosis. Unexpected deaths due to epilepsy were recorded in 19 (3%) cases.
Ferro et al.	2013	Purpose: Identified risk factors that might shape five QoL trajectories among subgroups of children. Findings: Baseline factors that increased risk for worse QoL included number of anti-seizure drugs, co-morbid cognitive and behavior problems, poor family functioning and parental depression.
Jacoby et al.	2011	Purpose: Linked QoL and clinical profiles in five trajectories in adults with new-onset epilepsy traced prospectively over four years.

		Findings: Persistent seizures correlated with poorer QoL scores suggesting underlying neurobiological mechanisms. At baseline between group differences were predictive of trajectories. Depression at baseline was associated with persistent seizures.
Neligan et al.	2011	Purpose: To understand the role of time for prognosis, used modelled heterogeneous patterns of remission and relapse in chronic refractory epilepsy among adults. Findings: Three pathways were identified: long-term remission, no remission, and intermittent periods of remission. Fewer anti-seizure drugs predicted better outcomes, although even after six anti-seizure drugs a small number of patients became seizure free. Early remission did not guarantee long term seizure freedom. Type of epilepsy made a difference.
Micallef et al.	2010	Purpose: Examined psychological outcome patterns in childhood onset temporal lobe epilepsy treated with or without surgery. Fifty-three subjects with epilepsy beginning before age 15 years were prospectively followed for 13 years. Mean age at last follow-up was 20 years. Findings: Identified four categories with distinct challenges. The most powerful predictors of category membership were personal sense of “cure”, self-reported increases in activity levels, and depression scores.

Appendix C: Philosophical Foundations: Key Points and Applications

Philosophy	Key points	Applications to the study
Pragmatism	- Pragmatism frames knowledge in terms of what a person finds useful to meet their needs; that which has been useful eventually becomes what the individual believes to be "true". - From a larger perspective of the collective, groups of people will find that which is most practical for solving human problems, i.e., that which makes a difference, to be that which is meaningful. - There is no one *Truth*, but many different interpretations give rise to a pluralistic reality. -Abduction is an essential concept within pragmatism and refers to the process by which useful explanations are developed.	- Illness experience in epilepsy is dynamic as it reflects the clinical and social course of disease. - It is possible that adaptive strategies are influenced by personal beliefs about the cause and course of epilepsy. - Treatment decisions are made based on what patients consider the most useful and what satisfies personal preferences. -Pragmatism has direct application because it emphasizes problem solving and refers to multiple perspectives.
Symbolic Interactionism (SI)	- SI is a perspective which assumes that people play an active role in shaping their world because of the way they think about it, and reflect upon it. - What is meaningful is constructed through processes of shared, social interaction. - Human interaction is about how we communicate and interaction relies on the symbolic meaning of words, actions, and objects.	- Personal illness narratives are reflected in how patients refer to seizures and epilepsy. - How people with refractory epilepsy see themselves (self) in relation to chronic seizures and their support network (others) may influence how disability is viewed, and the actions they take.
Constructivism	- What we know is constructed from personal realities i.e. how individuals interpret their personal worlds. - Knowledge is a human element that is socially constructed. - A constructivist approach examines the processes of 'what is happening' in the data.	- How patients with refractory epilepsy make complex health care decisions is under-explored. - Drawing upon what is useful (pragmatism) and meaningful (SI) in personal narratives facilitates the construction of deep, personal understanding of illness experience. - Researcher and participant work together to construct what is known.

Appendix D: Clinical Characteristics (N=35)

Age at interview in years: mean (range)	35.6 (18-68)
Duration of epilepsy in years: mean (range)	15.4 (2-44)
Age at seizure onset in years: mean (range)	20.2 (2-60)
	n (%)
Gender	
Male	15 (43%)
Female	20 (57%)
*Age at seizure onset: early versus late onset in years	
< 21y	24 (68.6%)
Mean (range)	12.7 (2-20)
≥ 21y	11 (31.4%)
Mean (range)	36.4 (26-60)
*Number of AEDs at interview	
1	3 (8.5%)
2	16 (46%)
3	13 (37%)
4 or more	3 (8.5%)
**Verbal Comprehension Index (VCI)	32 (100)***
Borderline (70-79)	3 (9.5)
Low average (80-89)	7 (21.8)
Average (90-109)	15 (46.8)
High average (110-119)	3 (9.5)
Superior (120-129)	2 (6.2)
Very superior (130 and over)	2 (6.2)

*Source: Medical records

**The Verbal Comprehension Index (VCI) measures verbal reasoning ability. This includes the ability to listen to a question and create a verbal response that expresses the person's thoughts. The index is a composite score of three tasks that measure word similarities, vocabulary and comprehension.

***Neurocognitive testing was not done for three participants.

Appendix E: Socio-Demographic Characteristics (N=35)

Characteristic	n (%)
Gender	
Male	15 (43)
Female	20 (57)
Ethnicity	
Caucasian	23 (66)
Hispanic	7 (20)
African American	4 (11)
Asian	1 (3)
Marital Status	
Never married	16 (46)
Married	13 (37)
Divorced or separated	6 (17)
Living situation	
Alone	3 (9)
With spouse and children	13 (37)
With children (no spouse)	3 (9)
With parents	12 (34)
With room-mate	4 (11)
Highest level of education	
≤ High School	15 (43)
Some college	9 (26)
Completed undergraduate degree	9 (26)
Completed graduate degree	2 (6)
Employment status	
Gainfully employed (full, part or self)	16 (46)
Unemployed	17 (49)
Retired	2 (5)

Appendix F: Examples of Conversational Interview Questions

1. You have been living with the seizures for a few years now. Can you tell me what it has been like for you to be living with seizures?

   Prompt: How do you understand getting seizures?

2. People use different words to explain the condition. Can you explain how you see the differences between epilepsy and seizures?
3. Can you tell me about some of the challenges you have had in coping with seizures?
4. What is it about the onset of the grand mal seizure that opened your eyes that something major is about to take place?
5. What made you decide at this time, that you needed to do more to treat the seizures?
6. How severe do you think your epilepsy is?
7. On a scale of one to ten, with one being most manageable, ten being the worst possible, where would you place your epilepsy?

   Prompt: Can you tell me why the number is not higher?

   Can you tell me why the number is not lower?

8. Supposing you were to meet a (age approximate to participant's seizure onset)-year-old person who is having seizures. What advice would you give that person?

   Prompt: What have you learned about living with seizures that has troubled you and that may help another person?

9. Many people we have talked to say that having to depend on other people causes anxiety and worry. What is it about having to rely on someone else that might cause anxiety for you?
10. If you were to get rid of the seizures, how do you think your life might change?
11. Can you describe the surgery that is going to be happening?

    Prompt: How do you feel about the surgery that is proposed?

12. How do you know the seizures are bad enough to warrant surgery?

Appendix G

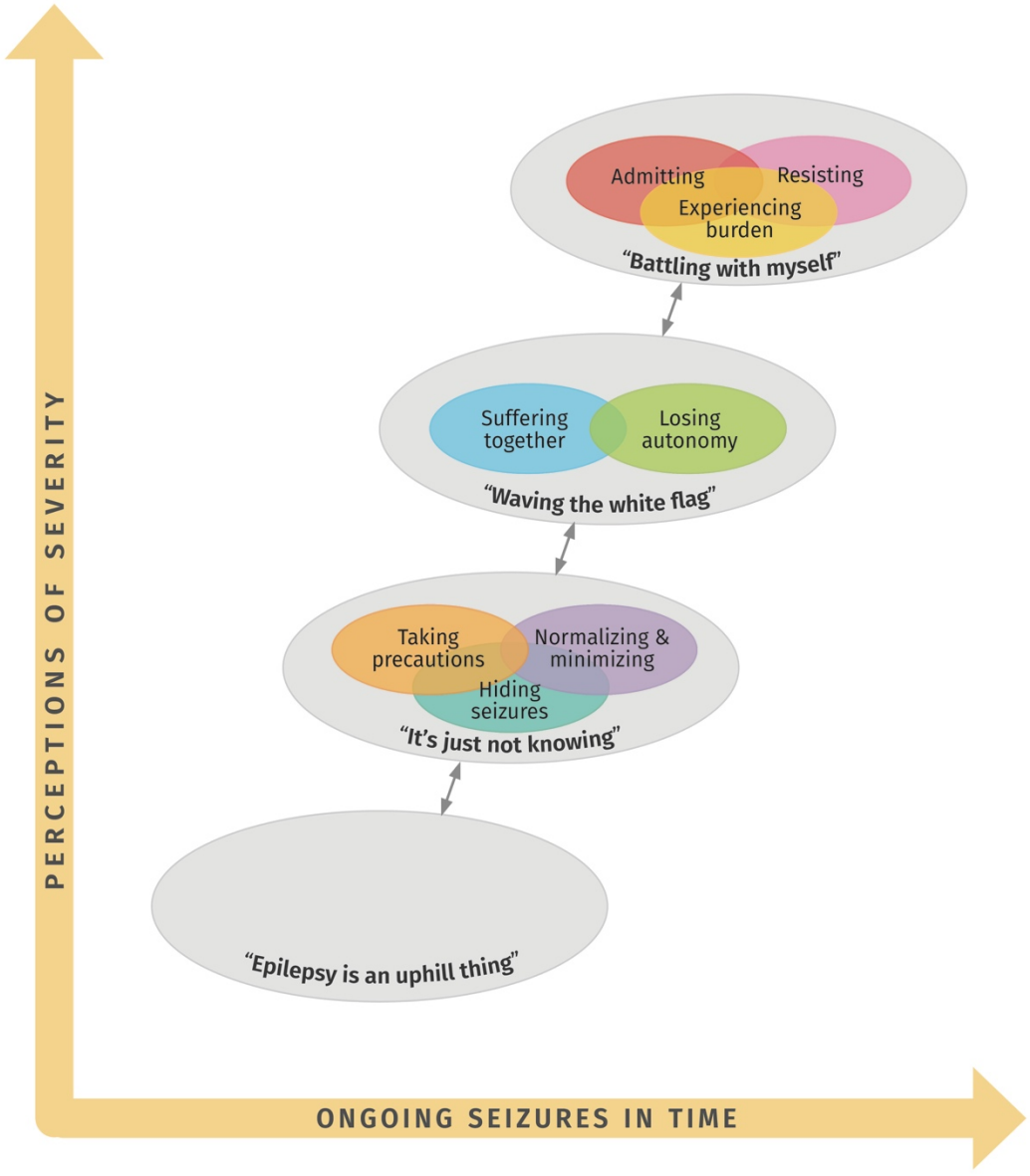

Figure 1. Perceptions of illness severity in adults with focal drug-resistant epilepsy.