Reciprocal burden: Adults with drug-resistant epilepsy reflect upon informal caregiver support

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A B S T R A C T

Purpose: To explore how a sample of people with uncontrolled seizures describe their experiences of receiving informal supportive care.

Methods: Using constructivist grounded theory, in-person, semi-structured interviews were conducted in 35 adults with drug-resistant epilepsy. These 20 (57%) women and 15 (43%) men were aged 18-68 years (mean= 35.6 years), with a range of verbal comprehension scores. The majority, 28 (80%) lived in nuclear family settings. Unpredictable seizures disrupted personal autonomy and generated unique challenges for everyday life. While supportive care was deemed necessary to survival, subjective reflections around the implications of care-receiving and caregiving, were ultimately experienced as mutually burdensome. Four dynamic and interactive dimensions revealed a mirrored, interactive perspective of what it meant to be a care-recipient: assuming responsibility; protecting and supervising; acknowledging reliance and setting boundaries. Care-recipience was not one-sided, but included actively hiding personal struggles as a way to shield the caregiver from the emotional and physical demands of caregiving. Relationship dynamics between caregivers and care-recipient played a major role in treatment decision-making. A clinically useful Theory of Reciprocal Burden resulted from our study.

Conclusions: This study adds an explanatory dimension to the concept of illness burden from the perspective of care-recipients. Relationship dynamics play a key role in patient-centered epilepsy care, with clinical implications for guiding supportive caregiving, fostering independence and promoting self-management strategies.

1. Introduction

Drug-resistant epilepsy (DRE) is a serious disorder characterized by unpredictable and intrusive seizures. Due to uncontrolled seizures, poor memory and cognitive deficits, family caregivers have to be relied upon for disease management and emotional support [1]. As a consequence, epilepsy is as much a focus of the lives of caregivers, as it is for those diagnosed with the condition [2-4]. Caregiving is frequently subtle and varied, both with respect to how carers identify with the role and the tasks they undertake. Caregivers themselves often do not identify as such, but assume that they are behaving as a loved one should [5,6]. In addition, cultural sensitivity plays a role in how the responsibilities of caregiving are shouldered [7]. Thus, labeling these informal caregivers is not simple, and encompasses various terms such as primary support persons [3], carers [6], caregivers [8,9], or family caregivers [10].

In general, providing care for those with chronic illnesses occurs in outpatient settings where families are relied upon to make health-related decisions and perform caregiving tasks. In DRE, caregiver-patient dyads are described as vital to illness management, yet the dynamics of caregiving, especially in adults with epilepsy, appear under-studied and focused mostly on the perspective of carers and their needs [8,11]. In a British study, attachment styles were explored to better explain the complexity of dyadic relationships in patients with seizures [4]. Patient distress was highlighted to significantly influence the quality of life of carers [4], supporting the recommendations that health care services be tailored to include the physical and emotional wellbeing of primary caregivers [11].

Studies in epilepsy about the role of caregiver-patient dyads highlight important differences between the perspectives of patients and primary caregivers relative to the nature of caregiving. Patients have been found to be more likely to express “resignation” around needing care and were more concerned about the impact of frequent medical crises than their caregivers [1]. Factors such as lower patient quality of life scores, higher numbers of anti-seizure drugs, and lower levels of caregiver education contributed to increased caregiver burden [8]. Additionally, the combination of intellectual disability and epilepsy, was found to carry greater disease burden for patient-caregiver dyads in comparison to the general epilepsy population [12], emphasizing distinct sub-group challenges for clinicians.

From the subjective perspective of caregivers, the negative impact of epilepsy on quality of life has been well described and clearly linked to caregiver burden [2,7,8,12,13]. Yet how the recipients of care perceive the personal meaning of care-recipience in everyday life appears under

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researched. We previously explored subjective perceptions of illness severity, and found that supportive care was spontaneously described as vital to navigating treatment decision-making in a severe and confusing illness [14]. Such references arose even when unprompted, and were powerfully laced across interviews drawing attention to a complex aspect of illness experience. Thus, as part of the parent study [14], our purpose here was to focus specifically on exploring personal perceptions of receiving informal care in patients with DRE to develop a clinically useful theory. The assumption at the outset of the research was that patient perceptions of caregiver support was a meaningful and perhaps vital component to comprehensive epilepsy care and treatment decision-making. For our study, the definition of caregiver included any family member, friend or partner identified by the person with epilepsy (PWE) as the one who witnessed the majority of seizures, and provided most of the informal care.

2. Methods

The methodology of constructivist grounded theory (CGT) was selected to study the human experience of living with a serious, chronic condition and develop a clinically useful theory [15]. We explored an emergent research question that became apparent during the analysis of a rich data set comprising the parent study [14].

2.1. Study design, recruitment and data collection

After satisfying institutional research board requirements, English speaking patients over age 18 years, with temporal lobe epilepsy (TLE), who were able to sign their own informed consents and who had not undergone previous epilepsy surgery were recruited at the specialized, tertiary level epilepsy center where they received treatment for DRE. Thirty-nine patients were offered participation, but four patients preferred not to be interviewed. The reasons four patients declined were not explored to avoid any perception of coercion, particularly since recruitment took place in the clinical setting. In-person, private interviews were conducted either during an inpatient video-EEG admission, in the patient’s hospital room (n = 16) or in the outpatient clinic soon after the consent for epilepsy surgery was signed (n = 19). A total of 51 interviews were conducted by a single, skilled interviewer (HP) with 35 participants lasting an average of 91 minutes each; the 16 second interviews gave additional opportunity to explore perceptions of a multifaceted illness. (See Table 1 for examples of semi-structured questions.) As unprompted descriptions about the significance of caregiver support unfolded in earlier interviews, constant comparison [16] led us to modify interviews to elicit deeper explanations supported by everyday examples. The audio-recorded interviews were professionally transcribed, checked for accuracy and de-identified. The software program, ATLAS.ti was used to manage the data, and to facilitate analysis [17].

2.2. Data analysis

Drawing upon the principles of grounded theory methodology, the analytic process embraced three levels of systematic coding undertaken by the authors who comprised the research team. Initial codes (first level) were sorted and labelled during the second level of abstraction to create focused codes. As codes were compared within and across participants one core category with two sub-categories became evident [18]. During the third level of abstraction, theoretical coding, we consistently drew upon reflexive memos and field notes, and diagrammed relationships to show how interpretations of the data were linked to the core concept [15]. Methodological rigor was upheld through an iterative process of analysis that included successive independent coding by the authors and regular team meetings to check interpretations [19]. In the case of differing opinions, consensus was reached through further discussion. The three members of the analytic team carefully deliberated between the core concept and the data, taking into account the context and salience of the interpretations. During this thorough analytic process, mutual burden consistently surfaced as the major category in the analysis.

3. Results

3.1. Sample characteristics

The study sample (N = 35) included 20 (57%) females and 15 (43%) males of which 23 (66%) self-identified as Caucasian. The average age at interview was 35.6 years (range 18–68). Seizures began before 21 years (range 2–20 y) in 24 (68%) participants, with the remainder beginning between the third to the seventh decades of life. Almost half had never married (16; 46%). Most participants (n = 28; 80%) lived with family members, except for 3 (9%) who lived alone, and 4 (11%) who lived with roommates. Most individuals (60%) also reported symptoms of anxiety and/or depression. Verbal comprehension scores reflected a sample with a range of verbal reasoning abilities. High school education had been completed in 15 (43%) participants, and half the sample reported being unemployed. See Table 2 for further details. The majority of caregivers were female (n = 24; 68.5%) however, the identification of this mother, wife, sister or daughter was not immediately evident, such as a teenager who lived with her mother yet identified her grandmother as the main support person, and an unmarried participant who leaned mostly on her young daughter.

3.2. Findings

While uncontrolled seizures signified a restricted future for both care-recipients and caregivers, our participants spontaneously acknowledged that their lives could not be lived without supportive care. Such supportive care provided a safety net that bridged cognitive deficits and met emotional, social and physical needs. While participants did not want to impose on others, and mostly required a caregiver to “simply be there”, they felt the impact of their epilepsy was not always clearly understood. Fulfilling the expectations of normal family roles, despite a challenging illness, was especially difficult for women and mothers (n = 11; 31.4%). A mother exemplified this inherent ambiguity of daily life, when she encapsulated her experience as follows: “We’re not alone, but yet we are alone in this.”

Overall, an appreciation for mutual burden lay at the nexus of perceptions of informal care. The dynamic nature of mutual burden reflected how participants imagined the experience of caregivers and what subjective interpretations of receiving care meant for the self. These mirrored perspectives served to explain a complex human response fundamental to illness experience and treatment decision-making. A process of systematic analysis led to an iterative Theory of Reciprocal Burden that represents multiple realities and diverse perspectives as illustrated in Fig. 1. The figure can be folded upon itself in many directions to demonstrate reflected views around the processes of receiving and giving care.

In the current study, the primary or main caregiver was identified and named as the family member who spent the most time at home, witnessed the majority of seizures, and was considered to be a reliable and trusted advocate. Caregivers were typically mothers, sisters, or spouses. Across ages and genders participants felt closer to mothers than fathers and several...
Table 2
Characteristics of participants with TLE (N = 35).

| Clinical characteristics |   |   |
|--------------------------|---|---|
| Age at interview in years: mean (range) | 35.6 (18–68) |
| Age at seizure onset in years: mean (range) | 20.2 (2–60) |
| Duration of epilepsy in years: mean (range) | 15.4 (2–44) |
| Gender n (%) |   |   |
| Men | 15 (43) |
| Women | 20 (57) |
| Age at seizure onset: early versus late onset in years |   |   |
| < 21y | 24 (68.6) |
| ≥ 21y | 11 (31.4) |
| Mean (range) | 36.4 (20–60) |
| Number of AEDs at interview |   |   |
| 1 | 3 (8.5) |
| 2 | 16 (46) |
| 3 | 13 (37) |
| 4 or more | 3 (8.5) |
| Side of epilepsy |   |   |
| Right | 16 (46) |
| Left | 11 (31) |
| Unclear | 8 (23) |
| Psychiatric co-morbidities |   |   |
| None reported | 14 (40) |
| Symptoms of depression and/or anxiety | 21 (60) |
| 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) |
| Demographic characteristics |   |   |
| 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 roommate | 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) |
| 1Health insurance provider |   |   |
| Private (through own or parent employment) | 25 (71.4) |
| State Health | 10 (28.6) |

Notes:

1 Source: Medical records.

2 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.

3 Neurocognitive testing was not done for three participants.

Source: Medical records.

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3.4. Dimensions of mutual burden

Four dynamic and interactive dimensions were highlighted within the two subcategories of receiving and giving care. In everyday life, perceptions of care-recipience were about acknowledging that reliance on others was necessary, but setting boundaries was essential to asserting an adult self. Perceptions of caregiving centered around assuming responsibility, protecting and supervising. In Table 3, selected, examples are provided for each dimension to illustrate intertwined and interdependent perceptions centered around mutual burden. Conceivably, quotations could overlap dimensions.

Acknowledging reliance: Participants relied upon caregivers to recognize seizures and bridge post-ictal memory lapses. Gaps in memory were particularly disruptive, leaving participants to describe themselves as simply “going through the motions of living.” Post-ictal confusion was described as a “state of mind” that deprived one of one’s thoughts leaving many to speak of the need to be calmed and protected until fully oriented. The risk of falling especially required caregivers to manage post-ictal behavior, and in general to “just keep an eye on me.” Although help from caregivers was essential to well-being, there was great...
concern that seizures were embarrassing and distressing for caregivers. Episodes of loss of awareness, especially convulsive seizures, made it particularly unsafe to be left alone with young children. These social limitations meant life was hard for everyone. Whilst sadness and distress were viewed as mutual burdens, participants were unanimous in their expressions of empathy for those caring for them and purposefully acted to reduce worry. As participants reflected on what it was like to depend on care, many spoke about simultaneously reassuring the caregiver.

**Setting boundaries:** The contribution of caregivers to medical care and treatment decision-making was emphasized and valued across interviews. Many participants spontaneously assumed responsibility for teaching others about epilepsy and dispelling stigma. Some described that society’s fear of seizures was transferred to a fear of the person with epilepsy. Being able to talk openly about the condition was described as vital to receiving caregiver support. However, setting limits on what information was shared was a strategy used to preserve personal dignity and protect the caregiver from worry. A professional woman explained:

My mother wants to take over, and wants to be able to fix the epilepsy, which she’s unable to. Nobody can fix this… at one point she asked if she could call my neurologist and ask him questions. I said, “No…I’m 34 years old. I can do this on my own”.

In order to cope, many participants admitted hiding or ignoring seizures and rejecting notions of personal ill-health. Irrespective of the duration of epilepsy, several participants expressed the importance of maintaining and improving personal health, for the sake of caregivers. Although the epilepsy was integral to self-identity and associated with limitations, the condition was not necessarily equated with disability. In setting personal limits around the consequences of chronic epilepsy, one professional said, “[Epilepsy] is who I am… I don’t look down on people who have conditions, or issues or disabilities. But I don’t want to be one of those people.”

**Assuming responsibility:** Caregivers were perceived to automatically assume responsibility for providing practical elements of health care, and relying upon them was accepted as inherent to living with seizures. Caregiving was viewed as time consuming and included having to learn about the disease itself. While participants gratefully accepted the need for assistance, having to depend on others was disruptive, burdensome and evoked feelings of guilt. Participants drew strength from the guidance and advocacy of caregivers that also served to reinforce a sense of self-worth. Caregivers “consistently worried” and remained concerned, unlike the transient nature of friendships. Across interviews, the importance of caregiver attendance at neurology appointments was mentioned, for the purposes of reporting on interval events and helping to remember the treatment plan. However, participants mostly desired active participation during clinic visits, and did not want to feel marginalized even when communicating was difficult. Personal maturity was questioned when participants felt excluded from health-related decisions.

**Protecting and supervising:** Seizures impacted the safety of everyday activities such as crossing streets, or going to public places. These risks often led to staying home. A teenager spoke about constant vigilance, saying, “If I am not being supervised by my parent then it’s my brother keeping an eye on me.” Participants disliked “being watched”, especially by siblings that were perceived to have an unclear understanding of the
Table 3
Mutual burden: Supportive examples of subcategories with dimensions.

| Subcategories                  | Dimensions             | Examples                                                                 | Illustrative links to mutual burden                      |
|--------------------------------|------------------------|--------------------------------------------------------------------------|----------------------------------------------------------|
| **Perceptions of receiving care** | Acknowledging reliance | “I have to wait for somebody to do it, somebody to take me over there... I have to wait because I have no choice.” [P’19] | Losing spontaneity in living                             |
|                                |                        | “I still feel I haven’t grown up because I live with my parents. I still feel like a little kid.” [P’01] | Not meeting expected milestones                           |
|                                |                        | “Sometimes I feel like a burden. I have to rely on my husband for everything I have to do. He has to take me to work. He has to pick me up. And then we have to go pick the kids up...” [P’31] | Describing frank burden                                   |
|                                |                        | “I don’t know how I would help the doctor find a solution because I can’t give him the information.” [P’31] | Relying on spouse to overcome personal helplessness       |
|                                |                        | “I felt... a sense of guilt because of all the medical things that I’ve had to pay for over the years. She [sister] wasn’t given the same opportunities to attend the schools that she wanted to go to.” [P’15] | Expressing guilt                                          |
|                                |                        | “She’s [mom] involved with all my medical. And that’s the way I like it.” [P’11] | Giving over control                                       |
| **Setting boundaries**         |                        | “The last thing I want to do is make my problems your problems. I don’t want anybody sad for me. Telling my whole family [about epilepsy surgery] and seeing them all break down was so hard.” [P’17] | Mutual caring and empathizing                             |
|                                |                        | “I’m already sad and upset for myself. So, her [mother] being upset on top of my upsetment turns into both of us being angry.” [P’10] | Perceiving shared sadness                                |
|                                |                        | “Treat me like I’m normal. Don’t look at me as... yes, I’m disabled, but don’t look at me that way.” [P’11] | Rejecting perceptions of disability                      |
|                                |                        | I don’t want to put my dad through stress. I want him to go to work and be calm. I don’t want him to have a bad life. I don’t want him to worry about me. ‘I’m sorry this [epilepsy] happened.” [P’27] | Limiting impact                                           |
|                                |                        | “If you don’t take care of your health, it’s going to decline. And the more it declines the less you’re going to be able to help your family.” [P’16] | Taking the role of the other                              |
|                                |                        | “I would like to think they [parents] kind of try to understand... that I do have the things that I need to try to control my emotions and all of that, as well with the medicine side-effects. I mean they have their moments to where it’s just like they don’t care. But I would like to think that they try to understand it.” [P’07] | Taking personal responsibility Sharing mutual challenges |
|                                |                        | “It’s not like you want this [seizures] to be happening... It’s what it is.” [P’02] | Yielding to perceived lack of empathy                     |
|                                |                        | “I comfort them, and I let them know that I’m strong enough that I can handle it [the epilepsy].” [P’08] | Expressing ambivalence                                    |
|                                |                        | “My parents are the ones taking me everywhere. I care about how they see my health... My mom has really tried hard to keep me healthy and for me not to get seizures.” [P’28] | Affirming self-sufficiency Maintaining health and well-being |
|                                |                        | “It’s very hard for us as epileptics to deal with what we’re dealing with and our family members to deal with it as well.” [P’20] | Empathizing                                               |
|                                |                        | “Doctors don’t listen to me until my mother shows up. And then they listen, because she’s kind of intimidating.” [P’10] | Advocating                                               |
|                                |                        | “My mother and my grandmother help me out a bunch with groceries and some of the bills, but I feel really bad having to have them do that.” [P’11] | Responding to dependency                                  |
|                                |                        | “My wife drives. I don’t watch my kids alone much, and that’s hard because my wife doesn’t get a chance to do a lot. If she wants to go shopping, my mom will watch the kids because, at any point, I could have something bad.” [P’05] | Acknowledging dependency as necessary                     |
|                                |                        | “She’s [daughter aged 9] very independent... She can cook, she can do dishes. She takes care of me. She’s done a lot to take care of me.” [P’03] | Validating perception of competent care giving            |
| **Perceptions of giving care**  | Assuming responsibility | “My parents are the ones taking me everywhere. I care about how they see my health... My mom has really tried hard to keep me healthy and for me not to get seizures.” [P’28] | Reciprocating care                                        |
|                                |                        | “It’s very hard for us as epileptics to deal with what we’re dealing with and our family members to deal with it as well.” [P’20] | Needing reassurance                                       |
|                                |                        | “Doctors don’t listen to me until my mother shows up. And then they listen, because she’s kind of intimidating.” [P’10] | Understanding mutual concerns                             |
|                                |                        | “My mother and my grandmother help me out a bunch with groceries and some of the bills, but I feel really bad having to have them do that.” [P’11] | Mutual dependency                                         |
|                                |                        | “My wife drives. I don’t watch my kids alone much, and that’s hard because my wife doesn’t get a chance to do a lot. If she wants to go shopping, my mom will watch the kids because, at any point, I could have something bad.” [P’05] | Empathizing with worry                                   |
|                                |                        | “She’s [daughter aged 9] very independent... She can cook, she can do dishes. She takes care of me. She’s done a lot to take care of me.” [P’03] | Expressing vulnerability                                  |
|                                |                        | “My parents are the ones taking me everywhere. I care about how they see my health... My mom has really tried hard to keep me healthy and for me not to get seizures.” [P’28] | Underestimating disability                                |
| **Protecting and supervising** |                        | “She [wife] has feelings for me, is looking out for my wellbeing, is responsive to things that happen to me, as I am for her.” [P’18] | Reciprocating care                                        |
|                                |                        | “They try to help me see how it’s not too bad and that they’re always going to be there for me and helping me.” [P’28] | Needing reassurance                                       |
|                                |                        | “I know my parents hate epilepsy because they also want me to do what I wanted to do, and that was to be a pilot... They hate epilepsy because they’re worried I might die, which yes, I could.” [P’12] | Understanding mutual concerns                             |
|                                |                        | “He’s [husband] going to be 70. So, we need to be healthy and able to go where we need to go and do what we need to do.” [P’21] | Mutual dependency                                         |
|                                |                        | (After describing serious threats to safety) “I can only imagine what my mom worries about every day.” [P’35] | Empathizing with worry                                   |
|                                |                        | “I’m a strong person. I just ask them to check on me. That’s all. I don’t want them to alter their lives or whole schedule, but you know, just check in.” [P’13] | Expressing vulnerability                                  |
|                                |                        | “It’s a catch-22 because they [family] don’t see me as disabled... they don’t know how hard it is.” [P’02] | Underestimating disability                                |

* Participant number
seizures. Care-recipient who characterized their lives as dependent and restricted, simultaneously empathized with support people who they viewed were saddened, disappointed and “tired out” from caring. Caregiving was about being protective, yet in reverse, participants employed strategies to shield caregivers. Seizures were hidden or minimized to limit caregiver stress, and many apologized for having the condition. Mutual suffering was described in relation to relentless worry about seizures that necessitated always remaining in close proximity to caregivers.

Many years of epilepsy frequently strained finances and caused unequal distribution of family resources. A woman in her 20s with seizures for 10 years said, “I need somebody to constantly take care of me… but [Social Security Administration] doesn’t want to give [disability insurance] to me.” Claiming social security benefits meant her mother could become her full-time “paid” caregiver. Integral to a supportive relationship was not assigning blame for having epilepsy; reassurance was needed, but not pity. Negative emotions and loss of personal dignity were woven throughout the data. Self-worth was impacted by not driving and was expressed as a significant challenge for the entire family.

Participant profiles were diverse with respect to the duration of epilepsy, age at onset and supportive needs, yet across the sample, common themes were evident in how caregiver support was regarded. While the need for a strong support system was generally acknowledged, supportive relationships depended on the generosity of others and impacted the ability of caregivers (often female family members and mostly mothers) to have a “normal” life. Overall, a mutually burdensome illness reflected shared suffering.

4. Discussion

Adults with DRE in this study perceived the role of family caregivers as critical to managing everyday life. Unpredictable episodes of altered awareness present unique challenges for daily life unmatched in other conditions. Studies in epilepsy [1-3,20] and other chronic illnesses [6,9] have explored the role of caregiving and quality of life of caregivers. We present a novel focus on what it was like for our participants to be the recipients of care, and how this impacted personal identity and treatment decision-making.

Unique to the present study is that the sample was relatively young (mean age = 35.6y), but had coped with chronic seizures for an average of 15.4 years. In contrast, studies of heart failure, included patients over 66 years [9,21]. As revealed in other studies [2,9,21] women in our study also provided most of the care. However, while these women included mothers, sisters and grandmothers, some participants relied on their very young daughters for day-to-day help and seizure safety. Episodic seizures created dependency and reliance on caregivers that was essential to survival. Everyday events, such as crossing a street alone or taking a bath in private, posed risks to safety that ultimately limited “normal” adult living, and evoked feelings of resignation around the need for supervision. Memory and cognitive deficits meant trusting caregivers to navigate both medical decisions, and life itself. Similar to other epilepsy studies, the lives of our participants and their caregivers were closely intertwined [3,11]. Although caring was perceived as natural to family relationships, participants were deeply empathetic to restrictions imposed on caregiver lifestyles, their constant worry and sadness. While various family members provided differing levels of comfort and support, and at different times, family roles were changed to accommodate disease limitations, specifically for participants who themselves were parents and/or spouses. Similar to the caregiving experiences of family caregivers in multiple sclerosis [8] and chronic heart failure [21], these changed roles were often discordant with expected social roles. Although the traditional labels of caregiver, or carer were less desirable, finding suitable alternatives was hard.

The overarching narrative of the current study included reflections around mutual illness burden (Fig. 1). Epilepsy was rarely managed alone, leading participants to reflect on multidimensional implications for themselves and their caregivers. While meeting pragmatic needs for day-to-day assistance mattered, our participants were acutely sensitive to the impact of their epilepsy on caregiver quality of life. The persistent need for care was perceived to cause resentment and stress for caregivers in our study as similarly reported by other authors [1,22]. To illustrate an awareness of illness impact on her family, one participant supposed, “They don’t hate me because I have epilepsy.” While supportive relationships depended on the kindness of others, participants expressed concern that caring had disrupted the “normal” lives of caregivers. Through a process of reciprocal exchange, the burdensomeness of epilepsy was mutually shared. Dependency impacted self-identity, yet all lives had to be adjusted to accommodate a range of needs related to safe and meaningful living. Mutual burden was about sharing worlds in ways that were often unwelcome. Relationships became changed to accommodate the needs for vigilant oversight, transportation and medical care. The illness narrative included acknowledgements of mutual sadness; a component of disease burden revealed only through personal accounts of illness in peoples’ own words [23].

Across participants, burden arose as an active and bi-directional experience. Witnessing seizures created mutual suffering described as “torture” for everyone. Yet, unpredictability left participants to describe themselves as tired and passive partners. Participants perceived that caregivers needed help and psychological support for the “burden” they carried. The desire to reverse disability and restore earlier competent selves was expressed by many whose epilepsy began later in adult life. Participants also attempted to alleviate burdensomeness by striving to express positive, independent self-images.

When contemplating the challenges of caregiving, participants did not refer to discussions with caregivers about illness impact, instead these were silent inferences throughout the interviews. While attempts to shield caregivers from the emotional, time and financial costs of illness were clearly evident, no mention was made of conversations with caregivers about these matters. Therefore, the work of shielding the caregiver from perceptions of burden was limited to silent conversations with oneself. These unspoken musings likely shaped perceptions of the self as care-recipient, to deepen understanding of complex, intertwined relationships also described as such by Walker et al. [3]. Grounded in the data, our Theory of Mutual Burden explains the intricate, reflexive processes around the meaning of care-recipience. Interpreting social cues was a process that focused upon what others were believed to think and feel about epilepsy, and to ultimately reveal that mutual burden existed at the center of caregiving relationships.

The disabling nature of epilepsy was often described as invisible to others even those living in close proximity. This meant that regardless of whether seizures were witnessed or not, others frequently had to be convinced that disability existed. Agency was important across participants regardless of whether the epilepsy was viewed as integral to personal identity. Living with invisible disability called for agency specific to educating others about epilepsy and to achieve autonomous treatment decision-making. In our sample, reducing burden on caregivers frequently motivated considerations for epilepsy surgery. This would appear to be a realistic assessment, as caregiver quality of life has been reported to mostly improve after epilepsy surgery [8].

The spontaneous conversations that occurred around the nature of caregiver support underscored our participants’ desires for open dialog. In an Irish study, the perception that epilepsy was negatively viewed by caregivers and society, contributed to hiding the seizures and denying the diagnosis [24]. Because negative perceptions have adverse treatment consequences, researchers have suggested that learned beliefs about illness should be considered in clinical practice [25]. The participants expressed pride in personal fortitude and wanted recognition for striving to be independent. Yet, along with a sense of indebtedness to caregivers, participants also described feeling misunderstood, lonely and unsupported, as powerfully expressed in the quote: “We are not alone, yet we are alone.”

4.1. Clinical implications

Table 4 is a summary of several useful, feasible clinical strategies based
on our Theory of Reciprocal Burden grounded in the narratives of the current sample. While we argue that constructive communication linked to tailored interventions can be developed to reduce perceptions of mutual burden and promote successful rehabilitation, a future study may explore how the perspectives of caregivers align with this theory. A logical next step is to understand the decisional impact of informal caregivers on the trajectory of disease outcomes, to meet the objectives of patient-centered epilepsy care outlined in the Institute of Medicine report (2012) [26].

4.2. Strengths and limitations

To our knowledge, this is the first study to explore what it means to those with DRE to be recipients of informal care. A study strength that contributes to analytic generalizability is the heterogeneity of illness experience across a sample that spanned ages, illness durations, and encompassed a range of verbal reasoning abilities. The current study provides a theoretical framework for understanding care-recipience with potentially transferable insights to other subsets of cases. While only adults with TLE were included, other epilepsy syndromes may have different clinical and quality of life implications, potentially limiting transferability of the findings. In addition, we did not explore the impact of seizure type and frequency on perceived burden. Caregiver demographics, such as age and employment status were not asked for and may be factors that influenced how participants perceived the nature of care recipience. Lastly, we recruited in a large, metropolitan academic center in the US, so this sample may not sufficiently reflect the lived experiences of those in other treatment settings.

5. Conclusion

A deep appreciation for reciprocal burden around needing and receiving care was inherent in participants’ views of supportive care. The caregiving relationship encompassed much more than simply meeting pragmatic needs, and powerfully shaped the ways recipients thought about themselves. Vigilant oversight was expressed as essential to survival, yet such dependency implied a deeply moral experience around living with severe illness. Writing from the personal experience of caring for his wife, Arthur Kleinman described supportive care as "far more complex, uncertain and unbounded than medical and nursing models suggest" [5 p.293]. We conclude that an ethical responsibility exists to address the multifaceted nature of illness burden in epilepsy that is foundational to the delivery of patient-centered, comprehensive health care across the spectrum of disease.

Declaration of Competing Interest

Sandra Dewar is a consultant for, and member of the Neuroscience Council Advisory Board of Neurelis Inc. Liza Ranit and Huibrie Pieters have no known conflicts of interest associated with this research.

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Table 4

| Dimensions                      | Findings                                                                 | Pitfalls                        | Communication strategies                  |
|---------------------------------|--------------------------------------------------------------------------|---------------------------------|------------------------------------------|
| Receiving care:                 | Most PWE rely upon informal caregivers.                                  | Opportunities for therapeutic dialogue are missed when discussion about supportive care is not invited. | Knowing the relationship dynamics between the support person and the patient may illuminate self-management approaches. |
| Acknowledging reliance          | Caregivers are critical to healing and adjustment.                       | Patients do not want to feel that they are a burden to the clinical team.                         | Clinician’s communication style contributes to patients’ sense of self-worth i.e. “I am worthy of being fixed.” |
| Receiving care:                 | Care-recipients shield caregivers from stress.                           | Conversations during clinic visits that do not actively involve the patient are perceived as disempowering. | Establish both shared and exclusive treatment goals. |
| Setting boundaries              | Guidance on how to manage caregiver emotions was requested.              | Short self-reported questionnaires in the patients’ own words maybe more useful than surveys in order to illicit what is individually meaningful. | Exploring protective boundary setting can be used to promote positive care-relationships. |
| Giving care:                    | Informal caregivers were integral to shared decision-making and treatment planning. | The identity of the primary caregiver may not be obvious, making it necessary to ask who fulfils this role. | Since families are often the first to observe deviations in health, patient-centeredness should include supportive caregivers as integral to the treatment team. |
| Assuming responsibility        | Relationships were complex, and centered around mutual burden, sadness and guilt. |                                | Approach the conversation with sensitivity. |
| Giving care:                    | Caregiver worry, fatigue and over-protectiveness can restrict independence and present as a source of family conflict. | Embracing the insights of caregivers may optimize limited clinic time. | Develop interventions that build educational and service resources within communities. |
| Protecting and supervising      | A health care system that only includes the physical care of the patient, negates a major dimension of holistic care. |                                | Assistance for caregivers should include social, financial, legal and ethical implications of caregiving. |

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