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IMPACT OF CAREGIVER BURDEN IN PAEDIATRIC EPILEPSY AT CHARLOTTE MAXEKE JOHANNESBURG ACADEMIC HOSPITAL, SOUTH AFRICA.

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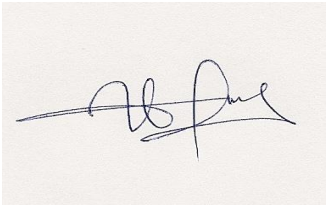


A Dissertation submitted to the Faculty of Health Sciences, University of the Witwatersrand, Johannesburg, in fulfilment of the requirements for the degree of Master of Science in Medicine

Johannesburg, 2018

DECLARATION

I, Dr Umar Abba Sabo, declare that this dissertation is my own work. It is being submitted for the degree of Master of Science in Medicine in the University of the Witwatersrand, Johannesburg. It has not been submitted before for any degree or examination at this or any other University.



Signature

19th February 2018

DEDICATION

To children with epilepsy around the world

ABSTRACT

Background: Chronic health conditions such as epilepsy may impose a high level of stress on the caregivers. The burden of epilepsy can cause significant dysfunction in the affected families resulting in a negative impact on the child's adaptation to the disease. This study seeks to evaluate the effects of caregiver burden on the health-related quality of life (HRQOL) of the caregivers and their family functioning as well as factors associated with high impact of the caregiver burden.

Methods: The participants consisted of primary caregivers who were involved in childcare for at least six months before study onset. Informed consent was obtained. One hundred and nine eligible caregivers recruited over a three months' period completed questionnaires providing information on their socio-demographic and epilepsy-related variables as well as paediatric quality of life (PedsQL) family impact module. High impact on HRQOL/ Family functioning was defined by Score below the inter-quartile range.

Results: The median HRQOL score of the caregivers was 46.3 (IQR = 31.3, 67.5) while the median family functioning score was 46.9 (IQR = 31.3, 71.9). In participants categorized with high impact, raw scores ≤ 31.3 were obtained for both caregiver burden and family functioning. The family functioning score correlated strongly with the HRQOL score of the caretakers, $\rho = 0.78$ and $p<0.001$. Multivariate analysis identified lower caregiver education level and a high seizure frequency as independent predictor of high impact of caregiver burden after adjusting for age of the caregivers and the number of anti-epileptic drugs taken.

Conclusion: The burden of caregiving for children with epilepsy in Johannesburg impacts negatively on family functioning. The burden of care was associated with high seizure frequency and lower level of caregiver education.

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NOMENCLATURE

AED(s) Anti-epileptic drug(s)

CMJAH Charlotte Maxeke Johannesburg Academic Hospital

CWE Children with epilepsy

HRQOL Health-related quality of life

PedsQL™ Paediatric quality of life inventory

QOL Quality of life

WHO World health organisation

CHAPTER ONE - INTRODUCTION

1.1 General Introduction

Epilepsy can be defined as the occurrence of two or more unprovoked seizures more than 24 hours apart (1). Epilepsy is the most common chronic brain disorder globally and affects people of all ages. More than 50 million people worldwide have epilepsy and 80% of them live in developing countries (2). Approximately 50% of people with epilepsy have their onset before the age of 5 years and 75% by the age of 20 years (3). A recent multicentre study in sub-Saharan Africa reported an age standardized prevalence of 8.1(7.5 – 8.7) per 1000 people for Mpumalanga, South Africa (4).

Epilepsy is characterized by a paroxysmal course that introduces the unique strain of unpredictability. It also has potentially significant cognitive, emotional, economic and social consequences that impact not only on those bearing the disease but also for their carers (5). Therefore, epilepsy does not only affect the individual, but it is a family problem.

The burden of care experience by families caring for children with epilepsy can be viewed as a multifactorial construct which includes emotional, psychological, physical, social and economic challenges.

Karakis et al (6) reported that epilepsy is associated with a modest degree of caregiver burden, and that caregiver burden has a negative impact on Health-Related Quality of Life (HRQOL) of the carers. While Nuhu and colleagues (7) found high caregiver burden in 52% of their subjects, Camfield et al (8) showed that the impact of paediatric epilepsy is negatively correlated to the

quality of life of both the children and their caretakers. They also found the impact to be associated with seizure frequency and number of anti-epileptic drugs (AEDs) taken.

Chronic health conditions such as epilepsy may impose a high level of stress on the caregivers. The burden of epilepsy can cause significant dysfunction in the affected families resulting in a negative impact on the child's adaptation to the disease. (9) Therefore, deficiencies in family cohesion, family adaptability, parent-child interactions, family conflict, and family problem solving skills can have significant impact on the family functioning in families of children with epilepsy (CWE). (10)

Therefore, periodic assessment of impact of caregiver burden and its determinants could contribute toward understanding the health care needs of the carers which will in turn improve the overall outcome of our management of childhood epilepsy.

The paediatrics quality of life (PedsQL ^{TM}) family impact module was developed to assess the impact of chronic medical conditions on the HRQOL of the caregivers and their family functioning. The PedsQL ^{TM} family impact module – items and scales were developed through focus groups, cognitive interviews and pre-testing measurement development protocol and the inventor's prior research and clinical experiences with children with chronic health conditions and their families. The PedsQL ^{TM} family impact module has internal consistency reliabilities exceeding the recommended minimum alpha coefficient standard of 0.70 for group comparison, with most scales approaching or exceeding an alpha of 0.90, recommended for individual patient/ subject analysis (9). These qualities make the PedsQL ^{TM} family impact module suitable for evaluating the HRQOL of the caregivers, the impact of childhood epilepsy on the family functioning and the factors associated with poor caregiver and family functioning.

1.2 Justification

Despite concerted efforts by paediatric neurologists to offer comprehensive epilepsy follow up services, the psycho-social and economic challenges faced by the caregivers are often neglected. Routine clinic follow-up visits focus mainly on seizure control, treatment compliance, school performance, behaviour problems and other comorbidities but do not include periodic assessment of caregiver burden and its impact on the entire family even in those with overt stress.

It is essential that the caregivers maintain their coping strategies because the psycho-social well-being of a caregiver is directly related to family functioning. If a family is not able to function, there may be a profoundly negative impact on the child's psycho-social adjustment to living with such a chronic condition (11).

Therefore, there is need for periodic evaluation of effects of caregiver burden and its implication on the family using systematic measures of their subjective experiences and perceptions. However, at the moment, there is little or no data on impact of caregiver burden in paediatric epilepsy in South Africa. It is therefore, hoped that the findings of this study will provide baseline data on this topic which may help to modify our epilepsy clinic protocol.

1.3 General Aim

- To determine the effects of burden of paediatric epilepsy on the carers and its impact on their family functioning, among caregivers attending paediatric epilepsy clinic at CMJAH.

1.3.1 Objectives

I. To assess the health-related quality of life of the caregivers of CWE attending paediatric epilepsy clinic of CMJAH using PedsQL ™ family impact module.

II. To determine the impact of burden of care of childhood epilepsy on family functioning using PedsQL ™ family impact module.

III. To identify factors associated with poor caregiver and/ or family functioning in paediatric epilepsy at CMJAH.

CHAPTER TWO - LITERATURE REVIEW

2.1 Caregiver Burden

A caregiver can be defined as a family member who is primarily responsible for providing everyday care of a child with epilepsy (6). While caregiver burden is defined as the physical, emotional/ psychological, social and financial problems that can be experienced by a caregiver (12).

Studies (5, 13) have shown that caregivers of children with epilepsy (CWE) suffer from fear and worry of cognitive deficits and learning problems that the child may experience as well as the unpredictability of the seizures. They also experience frustration, hopelessness, loss of self-esteem and confidence, guilt and anger (14). A common preoccupation of caregivers of CWE is fear of injury and death of the child because of the seizures. They also express concern about the future of the patients when they are too old or too infirm to care for the child. It has been reported that the level of parenting stress experienced by families of CWE was so high that professional intervention was needed.(5) Similarly, Chiou et al (15) found the level of parental stress is higher in parents of epileptic children than those with asthma. They proposed stigma, poor child adaptation and unpredictability of the seizures as the possible reasons for the higher stress. However, a contrasting finding was reported by Hoare and Kerley (16) that there was no significant difference in terms of psychiatric morbidity and marital adjustment problems between parents of children with epilepsy and the general population.

Epilepsy is highly stigmatized because of commonly held misconceptions that it is contagious or due to the patient being possessed by spirits or demons and other negative meanings attached to its outward manifestations. The negative perceptions as well as the demands of caring for such

children adversely affect the social life of the caregivers leading to poor personal care, poor sleep, little time for meals, withdrawal from out-door social activities, and/ or they stop inviting friends and relatives to celebrate social functions at their home (13).

Parents of CWE sometimes fear divulging their child's diagnosis to their friends and relatives because they experience a sense of shame, self-blame and rejection (17) and may consequently withdraw from their relatives and social circle which increases their risk of losing social support. Some caregivers have to cope with deteriorating marital relationships due to stress induced by blame, guilt, anxiety, tiredness and exhaustion. This could lead to poor communication, poor cohesion and integration, resentment and divorce.

The economic burden of paediatric epilepsy depends largely on the individual's setting, whether there is a functional medical insurance scheme, social security system or it is an out-of-pocket medical care system that operates. Reichmann et al (18) in a population-based cross-sectional study among 489 CWE and their caregivers reported a total direct cost of €1,619 per participant per 3 months. This was largely due to cost of hospitalization (€774, 47.8%), AEDs (€213, 13.2%) and ancillary treatments (€147, 9.1%). The total indirect cost was €1,231 in mothers, and €83 in fathers per 3 months. The high indirect cost was mainly due to loss of productivity in the mother. They concluded that paediatric epilepsy was associated with both high direct cost due to frequent in-patient admission and high indirect cost due to productivity losses in mothers.

In a related Nigerian study, Lagunju and co-workers (19) reported a total median annual cost of paediatric epilepsy of US$717 per patient per year (range of $155 - $21,900). Direct medical and non-medical costs accounted for 71.8% of the total mean annual cost. In-patient care and AEDs expenditure comprised 33% and 21.8% of the total mean cost respectively. More than half of the

families expended over 20% of their total family income on the care of the chid with epilepsy. The study concluded that the economic burden of childhood epilepsy in Nigeria is enormous with very high out-of-pocket expenses.

A similar study from Enugu, Nigeria (20) found a mean annual direct and indirect cost of US$162.6 and $82.3 respectively. Most of the direct expenditures were due to cost of AEDs (25.4% vs 35.3%) and investigations (48.7% vs 61.3%) as out-patients and in-patients respectively. All payments were made out-of-pocket due to lack of health insurance. These Nigerian studies were done in tertiary health facilities where only a limited proportion of CWE have access due to cost and poor referral services. Therefore, the findings may not apply to the general population.

2.2 Impact of Caregiver Burden on HRQOL of the Caregiver

2.2.1 Quality of life

The World Health Organization (WHO) has defined Quality of life (QOL) as the individual‘s perception of their position in life in the context of culture and value system in which she or he lives in relation to her/ his goals, expectations, standards and concerns.(21) While the Centre for Health Promotion defines QOL as the degree to which a person enjoys the important possibilities of her/ his life.(22)

2.2.2 Health-related quality of life

Health-Related Quality of Life **(**HRQOL**)** can be defined as individual’s perception of various aspects of their life that are affected by a particular medical condition (e.g. epilepsy) and its treatment. (21) Health-Related Quality of Life is multidimensional and for people with chronic

conditions such as epilepsy, it is often related to functioning in three main areas; physical, social and psychological domains.(23)

Nuhu et al (7) assessed level of burden of care cross-sectionally among caregivers of epileptics in Kaduna state, North-Western Nigeria. Using the Zarit Burden Inventory, and excluding carers of persons with comorbidities such as intellectual disability and affective disorders, they reported a high burden in 51.9% of their subjects.

Impact of caregiver burden on the HRQOL of the carers can be described in terms of physical, social and psychological/emotional domains.

2.2.3 Physical Impact

Paediatric epilepsy can affect the physical well-being of the caregiver negatively. Caregivers may experience chronic fatigue, sleep deprivation, lack of control over day-to day events, lack of time to complete daily tasks among other things. This has been linked to demands of caregiving.(24) Cottrell and Khan (25) studied the impact of childhood epilepsy on maternal sleep and socio-emotional functioning among 50 parents of preschool aged CWE. They reported that parents sleep about 4 hours per night, awaken at least 3 times per night to check the child, parental awakening was inversely related to parents' perception of their own quality of sleep, marital satisfaction and maternal health. It is a fact that disrupted night sleep is associated with excessive day-time somnolence, which could negatively impact on physical functioning, mood and sense of well-being. Some caregivers have identified the following as factors militating against their own health promotion; lack of time, lack of respite hours, and lack of qualified alternative care providers for the child and low prioritization of their needs. (26)

2.2.4 Social Impact

The demands of caregiving and child advocacy have been associated with restriction of leisure and social activities of the caregivers. This social restriction coupled with poor communication with their partners or spouses could result in relationship difficulty and/ or marital adjustment problems. There are reports that parents of CWE have a higher divorce rate than the general population. (21) Stigma can lead to withdrawal of the caregiver from their relatives and social circle with subsequent loss of social support. (13) Some parents have expressed their need to feel valued, acknowledged and understood by their extended families, friends and community.

The health of a child is managed in the context of the family, and mothers almost always assume that role probably because of societal ideology and/ or gender role assignment. (27) Therefore, good maternal adaptation to the child's diagnosis would be beneficial to the mother, the child and the family in general. Shore and colleagues (28) examined maternal adaptation to their children's epilepsy with the aim of investigating the association between maternal and child characteristics and maternal adaptation outcomes. The results of the study support that child internalizing behaviour problems; maternal learned helplessness and lack of maternal satisfaction with family relationship are associated with poor maternal adaptation to their child's epilepsy.

2.2.5 Psychological/ emotional Impact

Caregivers of CWE are worried about side effects of medication(s), cognitive deficits, the child's future and career, and they also experience anxiety. As one parent put it bluntly "witnessing one's child having a generalized tonic-clonic seizure is the most anxiety provoking experience one can ever have". (17) Parental perception of seizure control was found to be positively correlated with parental adjustment. (29) Maternal learned helplessness (low self-efficacy) is the

global perception that all actions are ultimately futile. (30) This could lead to depressive mood and poor adaptation to the child's epilepsy. (28) Prolonged perception of vulnerability and anxiety about the child's future could lead to greater psychological strain for the parents. (14)

Buelow et al (31) examined sources of stress among parents of CWE and mild intellectual disability. Parents reported stress related to uncertainty about future, communication with healthcare providers, changes in family relationships, interaction with the school and support within the community. Parenting stress, which is the type of stress that is uniquely perceived by parents and results from the demands inherent to being a parent, was found to be high among caregivers of CWE. (32)

Aytch and colleagues (33) investigated parental perceptions and found that parents needed support and information about medical, developmental, emotional and family issues related to coping. However, they discovered that there was difficulty in accessing the information. In a related study, McNelis et al (34) observed that inaccurate or incomplete information about epilepsy can interfere with appropriate management and adjustment to the condition for both parents and their child. They also suggested that there is a need for assessing caregiver's information and emotional support needs during the first consultation/ diagnosis and that ongoing assessment is also necessary because parents have continuing needs beyond the initial encounter with healthcare professionals.

A qualitative study by McNelis and colleagues (34) was aimed at exploring the concerns about seizures, need for information and support of CWE and their parents. A focus group discussion was applied to a purposive sample of school-aged children and their parents. A semi structured interview guide with open ended questions was used under the guidance of a trained group

leader. The concerns of the parents from the study include difficulties, struggles, and/ or problems navigating the healthcare system, being an advocate for their child, understanding the trajectory of the disorder, working effectively with the healthcare providers and the changes in family roles. They discussed the need for information and adequate time to process the new information and they wanted to be given information that built on their existing knowledge. They also expressed their need for emotional and medical support. Parents also had fears and concerns regarding the present condition of the child and the possible complication(s) that could arise in the future. The unpredictability of when or if the next seizure would occur is also of great concern to them. However, the study is limited by the social desirability of the response, interaction effect and the dominance of the vocal members of the participants.

In a related study, Saburi in Zimbabwe (35) reported the following as the top 6 stressors of caregivers of school-aged children with epilepsy: inability to get anti-epileptic drug(s), deep pain/ sadness caused by the child's seizures, caregiving which was falling predominantly on mothers, limited help from extended family, inadequate information on side effects of drugs as well as inadequate information on seizures. She also identified religious/ worship groups as the most commonly utilized community resource, while epilepsy support groups are the least patronized.

It is important to note that emotion-focused coping strategies such as wishful thinking, self-blame and avoidance are largely ineffective and only serve to exacerbate the distress. Meanwhile, problem-focused coping behaviours, such as cognitive restructuring, enhancing social support and information-seeking behaviour tend to lower parenting stress. (36) Spending time on oneself (away from the child), by engaging in independent activities outside the family,

as difficult as it may be, was reported to be invigorating. Working also helps the well-being of parents and energizes them for the caregiving role. (30)

2.3 Impact of Caregiver Burden on Family

Having a family member with epilepsy can affect the quality of life of the entire family negatively. The social and emotional toll of care, sometimes round the clock, can place financial and emotional strain on marriages and families, altering roles, relationships, and lifestyle. (37) Datta et al (38) studied 132 families in India who had children between the ages of 4-16 years with epilepsy using Impact of Paediatric Epilepsy on the Family Scale. Their exclusion criteria were having severe medical comorbidities and intellectual disability because a prior study had reported high burden among them. They found negative impact (high impact) in 42% of the families. Multivariate analysis revealed four factors that were significantly associated with the high impact; short duration of epilepsy, poor seizure control, AEDs polytherapy and high behavioural problem score on the child behavior checklist. They recommended monotherapy and early diagnosis as well as treatment of behavioural problems.

The impact of caregiver burden on family can be discussed under three broad categories; impact on emotional health of family members, impact on social and leisure activities and impact on employment and role expectation.

2.3.1 Impact on emotional health

Some family members appear to be more vulnerable to high psycho-emotional impact of paediatric epilepsy than others. Ramaglia and colleagues (14) examined the impact of idiopathic epilepsy on mothers and fathers in terms of strain, burden of care, worries and perception of vulnerability. It was a longitudinal study, data was collected shortly after diagnosis (T_0) and a

year later (T_1). The control group was matched for socioeconomic status and also for family composition, number, age and gender of children. However, the sample size was small (25 parents of epileptic kids and 27 parents of children with acute upper respiratory infections or diarrhea as control). Nevertheless, at T_0 parents of CWE showed higher levels of worries and perception of vulnerability than controls. Moreover, mothers of CWE sustained greater burden of care and exhibited higher levels of strain than the fathers. At T_1 strain and perception of vulnerability had decreased for all parents, while burden of care and worries remained stable. At T_0 and T_1 strain was associated with parents' perception of vulnerability and anxiety for their child future.

In another study, Aronu and Ojinnaka (39) investigated prevalence of psychiatric morbidity among parents of CWE in Enugu, Nigeria. They had a large sample, 308 parents of CWE (index parents) and 308 parents of children without epilepsy (as control parents). The controls were matched for age of the parents, family size, and socioeconomic status. They excluded parents with another child with chronic medical condition(s) as well as CWE plus neurological comorbidities. They reported significantly higher prevalence of psychiatric morbidity among the index parents (34.4% vs 22.1%). They also noted a higher prevalence among mothers in the index parents (49.4% vs 20.9%). The high prevalence of psychiatric morbidity was significantly related to degree of seizure control.

The unpredictable nature of epilepsy creates uncertainty about the future of the child. This affects parental coping ability as evidenced by increased level of stress, negative moods and impaired family functioning. (11) The stress in the family environment has the potential to disrupt parenting behaviours and erode parents' confidence in their ability to parent their child. (29)

Some families appear to be vulnerable to higher psycho-emotional impact of caregiver burden than others. Such families include parents of younger children, unmarried couples, parents of children with associated comorbidities and were found to have high (negative) impact of caregiver burden. (40) Lack of emotional and practical support, (41) loss of sleep and financial burden (42) were also associated with increased depression among caregivers.

There are reports that childhood epilepsy is more likely to be emotionally upsetting for siblings of CWE than other diseases. (43) Siblings of CWE also experienced fear, worry, anger and anxiety. Some expressed concern about their parents being exhausted and unable to care for them. There was a report that a quarter of siblings of CWE have behavioural problem from jealousy caused by parental attention on epileptic children. (16, 44) Siblings also complained about lack of written information about the condition for them. (45)

2.3.2 Impact on Social and Leisure activities

Previous reports have shown that families of children with epilepsy have more relationship and parenting difficulties and thus a more disrupted environment than families of children with other chronic illnesses such as asthma. (46) This could be attributed to the hidden, episodic and unpredictable nature of epilepsy, its potential for injury and death, frequent occurrence of comorbidities and associated stigma. (37)

Paediatric epilepsy appears to impose restrictions on family social and leisure activities. Studies have reported that parents of CWE spend less time outside home on recreational activities than controls, (42) low self-reported quality of life rating, and lack of time to pursue personal interests. (47) The factors identified to be associated with high social impact include inadequate support from extended family members, lack of support from outside the family unit, the need to

provide caregiving and lack of awareness about the resources available in the community. (37) Thompson and Upton (41) suggested that lack of family leisure activities could contribute to the high emotional impact experienced by such families.

2.3.3 Impact on employment and role expectation

Caregivers miss work due to caregiving responsibilities and the psycho-emotional difficulties may interfere with their ability to concentrate and focus at the workplace and hence reduce their participation in jobs that require high concentration.

2.3.4 Family functioning

Family functioning describes how family members communicate, relate and maintain relationships amongst each other and the way they solve problems. (48) After the diagnosis of epilepsy, families undergo adjustment processes, which could last for months to years, to get back to “normal life” which refers to the way they lead their lives before the onset of the epilepsy. (48)

The family as a unit can be at risk of poor communication, poor cohesion (low level of emotional bonding between family members) and poor integration due to the unpredictable nature of epilepsy as well as other stressors highlighted above. (30) It was reported that an increase in the number of stressors is associated with deteriorating family functioning. (31) Therefore, considering this body of evidence, management of childhood epilepsy will be most effective through family-centred caregiving.

2.3.5 Family-centred caregiving

This involves ensuring that parents/ caregivers/ families have ultimate control over decision making, treating parents respectfully and supportively and providing families with needed information. (49)

The components of this healthcare package include:

a) Enabling and partnership with the family/ parents.
b) Providing general and specific information about epilepsy for the child and the family.
c) Coordinated and comprehensive care for the child and the family including strategies for promoting well-being.
d) Respectful and supportive care including social support networks.

Strategies promoting well-being could be central if utilized efficiently and could be cost effective even in our resource poor settings. These strategies can target the processes and factors involved in adjustment, coping and resilience. Parents and families could be encouraged to use problem-focused coping behaviours, which involve active attempts to solve the problem directly related to the source of their stress. This coping strategy is associated with higher levels of parenting efficacy (maternal adaptation to their new parenting role). (36)

Resilience of a family refers to the ability of the family to maintain healthy family functioning, adapt to stressful life events and subsequently develop strengths and skills. (50) Resilience based health promotion intervention can assist families to identify their strengths, recognize the protective factors and resources they can utilize and build on within their family and the environment and provide opportunities to practice specific strategies to improve coping and family functioning. (51) Studies have shown that helping families to identify positive coping

skills, enhancing family functioning, and assisting them to access resources including social support results in positive patient and family outcomes. (52) Similarly, there is evidence that interventions targeting parents have positive outcomes for the child and family. This is due to parental well-being and family functioning having a significant impact on the child's health outcomes and coping with the epilepsy and sibling adjustment. (53) Finally, it has been suggested that the efficacy and acceptability of parents-based intervention is enhanced when it is done in group(s) as group interventions have additional benefit of opportunities for exchange of information, sharing of experience and mutual support. (54)

2.4 Health Related Quality of Life Assessment Tools

Health outcome assessment is defined as evaluation of health products, services or programs and the consequences of their use. (55) Health outcome evaluation comprises clinical, economic and patient-based outcomes. Health-related quality of life is generally accepted as the best measure of patient-based outcome.

2.4.1 Types of HRQOL measures

There are two broad categories of HRQOL instruments, Generic and Disease-specific instruments.

Generic instruments are designed to measure all important aspects of HRQOL. Thus, they can measure general health status across diverse samples, including screening healthy population for specific problems related to their health and well-being. They are also good for comparisons across interventions or conditions. They can detect differential effects on different aspect of health status. However, they are limited by inadequate focus on areas of interest and may not be responsive.

Disease-specific instruments aim to assess health status within a well-defined clinical sample e.g. asthma or cerebral palsy patients. Hence they are more sensitive to clinical changes in the designated patient group (are more responsive). (56) However, they are not useful for comparison across diverse population including benchmarking with a healthy population. (55)

The HRQOL instruments can also be classified into Discriminative and Evaluative instruments. Discriminative measures are used to differentiate between 2 patient groups at a point in time, while evaluative instruments measure how much the HRQOL has changed over time. (56)

2.4.2 Modes of administration

Health-related quality of life questionnaire(s)/ instruments can be administered by trained interviewer or self-administered.

The interviewer method has the advantages of maximizing response rate, having few, if any missing item(s) as well as minimizing errors of misunderstanding. However, it requires many resources including training of the interviewer and may reduce willingness to acknowledge problems.

Self-administration method requires minimal resources but has greater likelihood of low response rate, missing items and misunderstanding.

Other methods of administration include telephone interview, supervised administration and surrogate responders (proxy).

2.4.3 Attributes of HRQOL instruments

Health-related quality of life instruments has the following attributes;

a) Conceptualization which refers to the theoretical or empirical basis of the measurement.

b) Measurement properties (reliability/ responsiveness, validity and practicality).

Reliability/ Responsiveness

For HRQOL instruments, reproducibility means having a high signal-to noise ratio, and for discriminative instruments, the way of quantitating the signal-to-noise ratio is called reliability. (56) Reliability refers to how well a measure reflects true scores as opposed to error. It is determined via internal consistency, or how well each test item correlates with the scale of which it is intended to be part. As a general rule, internal consistency should have an alpha value of at least 0.70 for group comparison and 0.90 for individual comparison. (55)

For evaluative instruments, the method of determining signal-to-noise is called responsiveness which is simply defined as the instrument's ability to detect change. (56)

Validity

Refers to how well an instrument measures what it purports to measure. (55) Criterion validity is applicable when there is a gold criterion/ standard to compare. An instrument is said to be valid if the results correspond to those of the criterion standard.

Construct validity is a theoretically derived notion of the domain we want to measure. It involves comparison between measures and examines the logical relations that should exist between a measure and characteristics of patients and patient groups. The theoretical framework provides a basis for understanding the behaviour of the system being studied and allows hypotheses or prediction about how the instrument being tested should relate to other measures. Validity is strengthened or weakened when the hypotheses are confirmed or refuted. (56) Note that validity

is not an all-or-nothing quality. Most HRQOL instruments have some but none has perfect in all situations. (56)

Practicality

This refers to the test's usefulness in a real-world setting as well as the ease with which it is administered, scored and interpreted. (55)

Guideline for selecting HRQOL instrument:

i. Practical i.e. easy to administer, score and interpret.
ii. Excellent reliability/ responsiveness and validity. Evidence from peer-reviewed journal articles should be standard for evaluating such instrument properties.
iii. Utility in diverse paediatric populations.

2.4.4 Comparison of Impact of Paediatric Epilepsy Scale versus PedsQL Family Impact Module

The Impact of Paediatric Epilepsy Scale (IPES) is a brief, 11-item, validated instrument which contains scales measuring the child's HRQOL and parents' worry about the child's future. (57) It is disease specific with good practicality has a scale measuring whether the child's health affects his relationship with family and friends. However, it does not measure the HRQOL of the caregiver and details of the family functioning as a unit.

On the other hand, the PedsQL ^{TM} Family Impact Module is a multidimensional instrument that could stand alone or be easily integrated into the PedsQL measurement model. It is a modified generic measure comprising 6 scales measuring parent/ caregiver's self-reported functioning and 2 scales measuring caregiver reported family functioning. It has been used across diverse

paediatric population/ conditions ranging from those with medically fragile conditions to a community sample/ family. (9, 58) It has excellent internal consistency reliability and construct and criterion validity. PedsQL ™ Family Impact Module was validated in developing countries like Brazil where Scarpelli et al (59) found it to exhibit adequate properties regarding the reliability, internal consistency and validity of the construct. Thus, it was recommended for assessing impact of a chronic paediatric health condition on the HRQOL of caregivers and their family functioning. However, cut-offs defining groups who suffer high or low impact have not been generally described. From the theory of psychometric testing it is known that the actual values of scales are dependent on the population in which they are applied and hence cut-off values cannot easily be accepted from one population to another. (60) Previous analyses using the PedsQL scale used as cut-off values mean - 1SD (61) and mean - 2SD from a healthy control group (62) to define high impact.

CHAPTER THREE - MATERIAL AND METHODS

3.1 Study Site

The study was conducted at the Paediatric Epilepsy Clinic of Charlotte Maxeke Johannesburg Academic Hospital (CMJAH), a tertiary level teaching and referral hospital in the province of Gauteng, South Africa, where there are also referrals from other provinces as well as neighbouring countries.

3.2 Study Design

The study was a descriptive cross-sectional study.

3.3 Study Population

The study population consisted of caregivers of children with epilepsy (CWE), between the ages of one and 16 years who are attending the Paediatric Epilepsy Clinic at CMJAH. Review of the clinic attendance register over the past six months gives an average of 160 patients seen per month.

3.4 Selection Criteria

3.4.1 Inclusion criteria

- Caregivers of CWE between the ages of one and 16 years who have been caring for the child for at least 6 months.
- Consent for the study.

3.4.2 Exclusion criteria

- Caregiver(s) of children with severe to profound intellectual disability and/ or autism.
- Caregivers of children with other chronic medical condition such as congenital heart diseases or sickle cell disease.
- Caregivers whose families experienced a major life event in the preceding 3 months e.g. separation, loss of job.

3.5 Sample Size and Sampling Method

3.5.1 Sample size

One hundred and nine (109) caregivers were drawn from the Paediatric Epilepsy Clinic based on feasibility and time limitations.

3.5.2 Sampling method

The first ten eligible caregivers were recruited every clinic day (weekly) at the Paediatric Epilepsy Clinic at CMJAH for the study period (October – December 2015).

3.6 Data Collection

The socio-demographic characteristics of the caregivers (age, gender, religion, education, employment, housing and time spent caring for patient in hours per week) were recorded on a data collecting sheet (Appendix B).

The child's clinical information such as age of onset of epilepsy, duration of epilepsy, average number of seizures per month in the past 3 months, number of anti-epileptic drugs (AEDs), as well as EEG and neuroimaging were also recorded on a second data sheet. (Appendix B)

Due to the non-availability of an epilepsy specific caregiver questionnaire to assess the impact of caregiver burden, the Paediatric Quality of Life (PedsQL ^{TM}) Family Impact Module (9) was administered to all participants. The PedsQL ^{TM} Family Impact Module (Appendix C) is a multidimensional instrument developed to assess the impact of chronic medical conditions on the caregivers and their families. It comprises of;

A.) Six subscales measuring caregiver's self-reported functioning:

i) Physical Functioning (6 items)

ii) Emotional Functioning (5 items)

iii) Social Functioning (4 items)

iv) Cognitive Functioning (5 items)

v) Communication (3 items)

vi) Worry (5 items)

B) Two subscales measuring caregiver's reported family functioning;

i) Daily Activities (3 items)

ii) Family Relationships (5 items)

The scale had five Likert response options, 'never', 'almost never', 'sometimes', 'often' and 'almost always' (corresponding to scores of 100, 75, 50, 25 and 0 respectively). The interpretation of the scale was such that higher scores indicate better functioning (less negative impact/ less impact). The PedsQL™ Family Impact Module Total Functioning Score (Total score) is calculated as the sum of the 36 item scores divided by the number of items answered.

Two other scores can also be obtained from the instrument; the caregiver HRQOL Summary Score and the Family Functioning Summary Score. The caregiver HRQOL Summary Score assessed the impact of epilepsy on the health-related quality of life of caregivers. The score was

computed as the sum of the 20 item scores on the Physical, Emotional, Social and Cognitive Functioning Subscales divided by the number of items answered in these subscales.

The Family Functioning Summary Score assessed the impact of epilepsy specifically on family activities and relationships. The score was obtained from the sum of the 8 item scores on the Daily Activities and Family Relationships Subscales divided by the number of items answered in these subscales.

The data sheet and the PedsQL ™ Family Impact Module questionnaire were administered by trained interviewers, the researcher and his assistant, a retired nurse, who is fluent in the most widely spoken local languages.

3.7 Operational Definitions

i.) Epilepsy is defined as occurrence of two or more unprovoked seizures more than 24 hours apart. (1)

ii.) Seizure frequency is categorized into zero seizure frequency (0 seizures / month), low frequency seizures (1-4 complex partial, 1 generalized seizures or 1-20 partial/ myoclonic/ absence seizures per month) and high seizure frequency (≥5 complex partial, >1 generalized seizures or >20 partial/ myoclonic/ absence seizures per month). (57, 63)

iii.) The number of anti-epileptic drugs (AEDs) taken by the children was classified according to the convention as no AED (0 AED taken), monotherapy (1 AED taken) and polytherapy (≥2 AEDs taken).

iv.) Social Support is defined as the support accessible to an individual through social ties to other individuals, groups and the larger community. (64)

v.) As no universal cut-off values for defining high and low impact groups based on the PedsQL scale are available, the 25th and 75th percentiles of the distribution of the current data were used. Negative/ high impact/ poor family functioning is defined as the HRQOL/ Family functioning/ Total functioning score below the interquartile range (in the lower quartile).

3.8 Statistical Analysis

The mean scores for the 3 summary scores of the PedsQL ™ Family Impact Module for each participant were calculated according to the developer's guidelines. (65) Missing items were also handled according to these guidelines. (65) Numerical data was described with mean values and standard deviation (SD) when symmetrically distributed and with median and inter-quartile range (IQR) when skewed. Categorical data was presented as frequencies and percentages. Bivariate associations between characteristics were investigated using unpaired t-tests, one-way Analysis of Variance, Chi-square tests, Spearman's rank correlation, and non-parametric Kruskal-Wallis and Mann-Whitney U-tests. The health-related quality of life score of the caregivers and the family functioning score were categorised using the quartiles of their distribution. The new variables were compared using a Chi-square test.

Multivariate linear regression analysis was used to identify independent associations between characteristics of the caregivers or the children with the total functioning score of the caregivers.

Categorical variables were dummy coded for this analysis. All characteristics were initially investigated as independent associates. After a model had been identified, remaining variables not in the model were assessed for confounding which was identified when estimates changed by 5% or more. The presented model was adjusted for confounding effects.

All statistical analyses assumed a p-value less than 0.05 to indicate statistical significance. Analysis was performed using SPSS (IBM SPSS, version 23).

3.9 Ethics and Consent

Ethical clearance was obtained from the human research ethics committee of the University of Witwatersrand (clearance certificate number **M150658** as Appendix D).

Informed consent was also obtained from the participants. A sample of the consent form can be viewed in Appendix A.

CHAPTER FOUR - RESULTS

4.1 General Characteristics of the Caregivers and CWE

The mean age of the 109 participants was 37.9 years (SD = 9.1) (Table 4.1). The majority (62.4%) of caregivers had completed high school. The median number of years the epileptic children had lived with epilepsy was 5 years (IQR = 2.5, 8). Less than forty percent of the children (39.4%) had zero seizure frequency during the previous 3 months.

The median HRQOL score of the caregivers was 46.3 (IQR = 31.3, 67.5), ranging from 1.9 to 95. The median family functioning score was 46.9 (IQR = 31.3, 71.9), ranging from 3.1 to 100. The mean total functioning score of the participants was 48.0 (SD = 23.6), with a range of 2.8 to 95.7 (Table 4.1).

Table 4.1: Description of characteristics of 109 caregivers and their epileptic child

Characteristic	Description
Caregiver & Family	
Age of caregiver, mean (SD)*	37.9 (9.1)
Level of education of caregiver, n (%)	
Primary school	18 (16.5%)
Junior secondary school	23 (21.1%)
High School	48 (44.0%)
Diploma	12 (11.0%)
Graduate higher education	8 (7.3%)
Caregiver employed, n (%)	46 (42.2%)
Social support received, n (%)	
None	10 (9.2%)
Government	48 (44.0%)
Family or friends	35 (32.1%)
Government and family or friends	16 (14.7%)
HRQL score of caregiver, median [IQR]**	46.3 [31.3, 67.5]
Family functioning score, median [IQR]**	46.9 [31.3, 71.9]
Total functioning score, mean (SD)*	48.0 (23.6)
Child with epilepsy	
Number of years lived with epilepsy, median [IQR] **	5 [2.5, 8]
Seizures frequency over last 3 months, n (%)	
None	43 (39.4%)
Low frequency	33 (30.3%)
High frequency	33 (30.3%)
Number of antiepileptic drugs taken, n (%)	
None	2 (1.8%)
1 (monotherapy)	30 (27.5%)
2 or more (polytherapy)	77 (70.6%)
Co-morbid condition, n (%)	74 (67.9%)

*SD = Standard deviation; **IQR = inter-quartile range

4.2 Impact of Caregiver Burden on Participants' HRQOL and Family Functioning

The high and borderline impact percentages were by the design of the study. In participants categorized with high impact of caregiver burden raw HRQOL scores < 31.3 were obtained (Table 4.2). Similarly, Table 4.3 displayed raw family functioning scores of < 31.3 in those with poor family functioning (high impact) attributed to their caregiving role.

Table 4.2: Impact of Caregiver burden on the participants' HRQOL

HRQOL Score Quartiles	HRQOL Raw Score	No of Caregivers (%)	Impact of Caregiver Burden (Level of Functioning)
Lower Quartile	1.9 – 31.2	28 (25.7)	High impact (Poor functioning)
Middle Quartiles	31.3 – 67.4	55 (50.5)	Borderline impact
Upper Quartile	67.5 – 95.0	26 (23.8)	Low impact (High functioning)

Table 4.3: Impact of Caregiver burden on the participants' Family Functioning

Family Functioning Score Quartiles	Family Functioning Raw Score	No of Caregivers (%)	Impact of Caregiver Burden (Level of Functioning)
Lower Quartile	3.1 – 31.2	29 (26.7)	High impact (Poor functioning)
Middle Quartiles	31.3 – 71.8	54 (49.5)	Borderline impact
Upper Quartile	71.9 – 100.0	26 (23.8)	Low impact (High functioning)

4.3 Correlation of Family Functioning and Caregivers' HRQOL

The family functioning score of the participants was strongly correlated with the HRQOL score of the caregivers (Spearman's rank correlation $\rho = 0.78$; $p<0.001$), (Figure 4.1). Of the 28 caregivers with a HRQOL score in the lower quartile, 18 (64.3%) also had a family functioning score in the lower quartile. Of the 55 participants with a HRQOL score in the middle quartiles, 41 (74.5%) also had a family functioning score between the 2nd and 3rd quartiles. Of the 26 caregivers with HRQOL score in the upper quartile, 21 (80.8%) also had a family functioning score of life in the upper quartile.

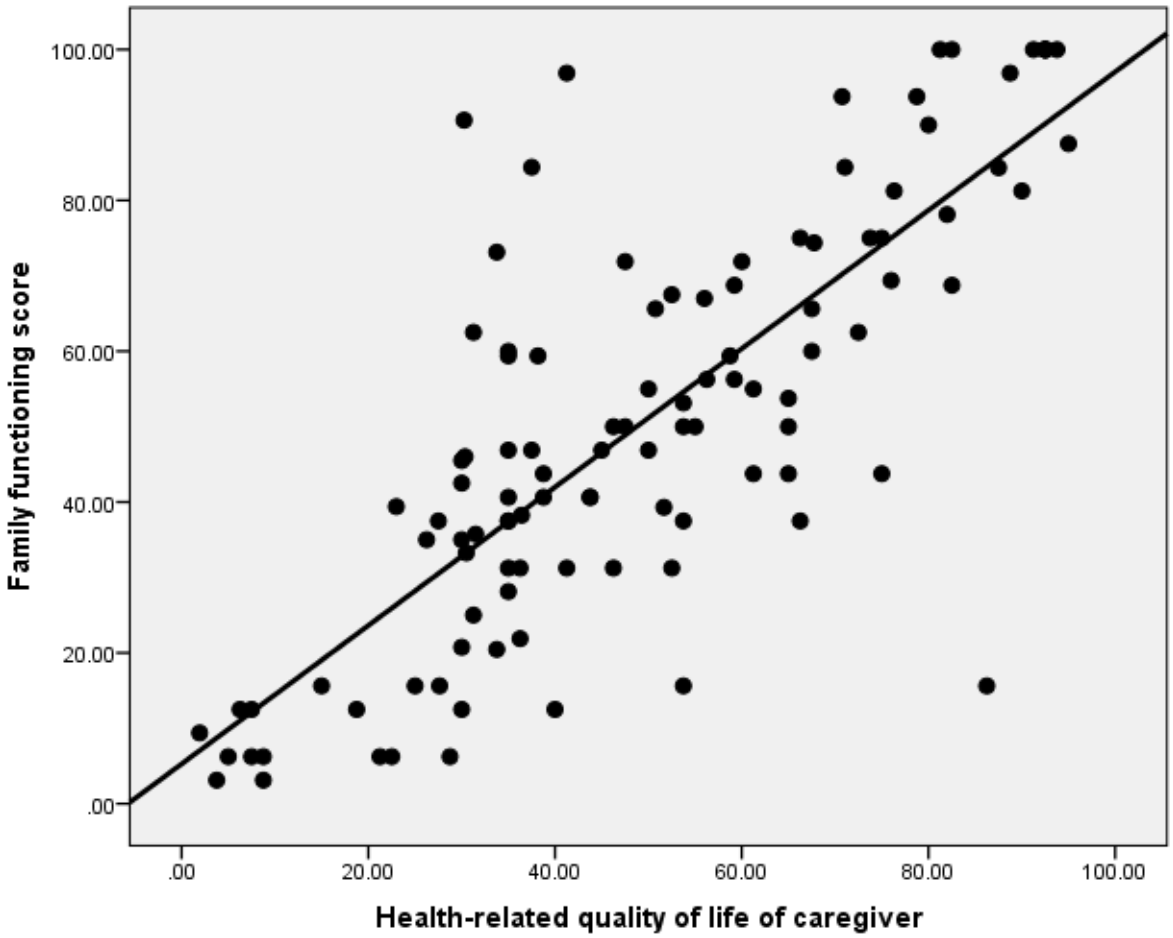


Figure 4.1: Correlation of Family Functioning and HRQOL scores of the caregivers.

4.4 Factors Associated with High Impact of Caregiver Burden

The caregiver's educational status, seizure frequency and number of AEDs taken were associated with the HRQOL score of the caregiver ($p<0.001$), family functioning score ($p<0.001$, $p=0.001$, $p<0.001$, respectively), and the total functioning score of caregivers ($p<0.001$,). For all three scales, higher levels of education showed higher scores (lesser impact), while a high seizure frequency or multiple AEDs taken were related to lower score (higher impact). The family functioning scores were highest (least impact) when social support was provided by family and friends (median 59.4) or the government and family and friends (median 60.9), and lower (higher impact) when social support was provided by the government alone (median 41.6) or no support was provided (median 48.4), ($p=0.028$). The remaining caregiver's and child's characteristics did not have significant association with the three scores.

Multivariate linear regression analysis confirmed level of education of caregiver and seizure frequency in the previous 3 months as independent predictors for the total functioning score of the caregivers (Table 4.4). A caregiver with a diploma or higher level of education had significantly higher total functioning scores (mean 64.4) compared to caregivers with primary school education only (mean 30.5, $p<0.001$). The total functioning score for caregivers was significantly lower in those with a high seizure frequency in the previous 3 months (mean 32.7, $p<0.001$). Both level of education and seizure frequency showed a dose-response relationship with the total functioning score of the caregiver.

Table 4.4: Multivariate linear regression of factors associated with high impact of caregiver burden.

Characteristic	Sample size n=109	Mean total functioning score (SD)**	Standardised coefficient	p-value
Caregiver level of education				
Primary school*	18	30.5 (18.7)		
Junior or high school	71	47.8 (22.8)	0.258	P=0.015
Diploma graduate or higher	20	64.4 (19.0)	0.425	P<0.001
Seizure frequency in the last 3 months				
Zero frequency*	43	60.8 (23.1)		
Low frequency	33	46.6 (19.7)	-0.263	P=0.003
High frequency	33	32.7 (18.2)	-0.441	P<0.001

*Reference category; **SD = standard deviation

4.5 Description of Health-Related Quality of Life, Worry, Communication and Family Functioning Scales by Seizure frequency

Health-related quality of life, "worry" and "communication" scales, as well as family functioning scales scores of caregivers consistently declined from higher average values in the strata of children with zero seizure frequency during the preceding 3 months, to lower average values in the strata of children with low seizure frequency, to lowest average values for children with high seizure frequency during the last 3 months (Table 4.5). All comparisons of HRQOL, "worry" and "communication" scales, and family functioning scales scores of caregivers between the three groups of children were statistically significant ($p<0.05$) using non-parametric Mann-Whitney U-tests.

Table 4.5: Description of HRQOL and family functioning scales scores of caregivers of children with poorly and well-controlled epilepsy.

Scale	Children with Zero seizure frequency during last 3 months (n=43)		Children with low seizure frequency during last 3 months (n=33)		Children with high seizure frequency during last 3 months (n=33)	
	Median [IQR]^	**Mean (SD)^^**	**Median [IQR]**	**Mean (SD)**	**Median [IQR]**	**Mean (SD)**
HRQOL scores of caregivers						
Total score	60 [45, 81.3]	61.2 (23.0)	50 [35.7,65.6]	50.2 (21.9)	31.5 [24, 38.8]	32.8 (17.5)
Physical	60 [41.7, 80]	59.4 (22.1)	50 [35.5, 70]	50.4 (22.4)	30 [20, 40.8]	31.7 (18.7)
Emotional	55 [40, 75]	56.4 (23.1)	40 [30, 60.75]	44.7 (20.1)	30 [22.5,34.2]	29.6 (17.9)
Social	64.2 [50, 87]	64.5 (23.7)	50 [35.8,65.2]	51.2 (22.2)	35 [25, 41.9]	34.0 (18.1)
Cognitive	68.8 [45, 88]	64.6 (25.3)	50 [37.5, 75]	54.5 (24.6)	35 [29.9,42.5]	36.3 (17.4)
Worry	50.5 [36, 70]	51.9 (23.1)	35.4 [20, 54.8]	38.5 (20.1)	25 [12.6, 35]	29.2 (24.6)
Communication	60.4 [40.4, 80]	60.4 (26.3)	41.7 [31.3,62.5]	45.2 (20.4)	30 [15, 41.8]	33.5 (24.7)
Family functioning scores						
Total score	65.6 [42.5,90.6]	65.0 (27.0)	46.9 [15.6,71.9]	47.9 (27.8)	35 [18.0,48.4]	34.2 (18.9)
Activity	56.3 [33.8,87.5]	60.4 (27.6)	37.5 [14.8,66.5]	42.6 (27.1)	30.5 [12.3,38.5]	28.3 (16.9)
Relationship	70.8 [47.5, 100]	69.6 (27.2)	55 [20.5, 75]	53.5 (28.5)	40 [20.5, 55]	38.2 (20.6)

^IQR = Inter-quartile range; ^^ SD= Standard deviation

CHAPTER FIVE - DISCUSSION

5.1 Discussion

This study was undertaken to determine the impact of caregiver burden on their health-related quality of life and family functioning as well as to identify factors associated with a high impact of caregiver burden.

There is limited number of studies that reported on impact of childhood epilepsy on the HRQOL of the caregivers, let alone the raw scores of the high impact group. This study found low HRQOL raw scores in those caretakers with high impact of caregiver burden. This is the first time such raw values are recorded in South Africa. These values could be used as guide for clinical practice or in research, since cut-off values from another clime cannot easily be transferred to our population. However, using a different assessment measure [Short-Form health survey (SF-36)], Lv et al in Beijing (47) reported significantly poor HRQOL scores among parents of CWE compared to those of healthy children. It is important to note that some caregivers may become isolated from their extended families and friends due to their caregiving role and this may limit their ability to pursue their own interests and hence their poor HRQOL. (66) Similarly, parental beliefs and attitude concerning epilepsy as well as the quality of parent-physician relationships were significantly associated with the HRQOL of the caregivers. (67)

In the current study, participants' families that were poorly functioning (high impact) due to the burden of care also recorded low raw family functioning scores. To the best of my knowledge, no previously published studies have reported raw scores of poorly functioning families to allow for comparison. Nevertheless, this result is an indicator of high burden of caregiving in paediatric epilepsy. However, there are studies that compared family functioning of CWE versus healthy

children. For instance, Tatzer et al (68) found 26% of the families of CWE were malfunctioning compared to 6.5% of those with healthy children. Our instrument was designed to assess how families function effectively as a unit which is similar to the formal social science measures of family functioning such as family assessment measure III (FAM-III) used by Tatzer et al. (68)

My data displayed that in those with high burden of care, 64.3% also had poor family functioning. This means there was a very strong positive correlation between the caregivers' HRQOL and their family functioning. This merit further exploration to determine if low caregiver HRQOL score could be used to predict poor family functioning and vice-versa. However, Bemister et al (69) found a spearman correlation coefficient of 0.86 between the HRQOL and family functioning of caretakers of children with perinatal stroke. But their value has exceeded the recommended cut-off point of 0.80 by Sweet and Grace-Martin for multicollinearity.

This study found that the higher the educational level of the caregiver, the lower the impact of paediatric epilepsy on both the caretakers and their families. Education is a resource that aids in acquisition and processing, in addition to facilitating organization. (28) A low level of caregiver education had been previously reported among families negatively impacted by childhood epilepsy. (15, 70) Judge (71) found an association between maternal education levels and the families effort to be active and innovative when their child has a disability. High caregiver education could also help change parental beliefs and attitudes concerning epilepsy, the perception of stigma as well as facilitate access to educational materials and support groups through social media networks as well as enhance parent-physician relationship.

High seizure frequency was significantly associated with high impact of caregiver burden on the carers and their families in the current study. This finding is congruent with previous studies. (25, 38, 39, 57, 70) and could be due to many reasons:

i) Caregivers of children with frequent seizures experience restriction with respect to time spent with their spouses, friends or time to develop their personal interest. (42, 47)

ii) Uncontrolled seizures may severely affect the employment of the carer due to frequent calls from the school when the child fits, which may lead to loss of job, less promotion and economic difficulties.

iii) Intractable seizures increase the risk of injury, behaviour problems, mood disorders, attention and cognitive deficits leading to high parenting stress. (66)

iv) Frequent seizures also increase the risk of discrimination and stigma at work and in the social setting. (47)

v) Parents have described witnessing their child's seizures as the most anxiety provoking experience. (17)

In addition, I found caregivers of children with a high seizure frequency had significantly lower HRQOL scores in all scales compared to those with a low seizure frequency, while those with zero seizure frequency had the highest HRQOL scores. Lv et al (47) reported similar findings (of significant difference) in all scales between carers of the groups depending whether there was poor or good seizure control. Mothers of children with intractable seizures have a high level of parenting stress and nearly two-thirds were found to be in the clinical range of Total Stress (scores above 85th percentile in both child and parent domains) on the parenting stress index.

(66) Moreover, the caregiver's satisfaction with seizure control has been correlated with good parental adjustment in families of CWE. (72) An interesting finding by Austin et al was that parental perception of seizure control was a better predictor of parental adjustment than the level of seizure control itself. (72)

Although the number of AED(s) taken was significantly associated with total caregiver functioning score, multivariate regression confirmed that it was not an independent predictor of high impact of caregiver burden, but rather has a confounding effect on seizure frequency. This augments evidence from a previous report that no significant direct relationship exists between parental stress and polytherapy. (73) On the contrary, Datta et al (38) found an independent association between polytherapy and high impact of paediatric epilepsy on the family even after controlling for behavioural problems. However, it is important to note that children with intractable seizures are more likely to be on polytherapy and these drug resistant seizures are more likely to be responsible for the high impact of caregiver burden than the number of AEDs taken. Nevertheless, inappropriate combination therapy is known to cause behavioural and psychiatric problems which can impair the functioning of the carers and their families. (74) Our participants were selected from a tertiary health facility where such errors are not prevalent. Moreover, AEDs are given free to all registered patients, which obviate the impact of cost of drugs on the caregivers.

The current study showed that family functioning scores were highest when social support was provided by family and friends with or without a government social grant and scores were at nadir when only government social grant was available. Social support is a valuable resource that is utilized by families caring for a child with disability. (71) In epilepsy research, social support was found to be associated with positive parental coping, (72) improved HRQOL of caregivers

(31) and decreased child behavioural problems. (75) Parental coping efforts directed at seeking informational and emotional social support are significantly associated with greater family-confidence strength. (71) Previous data have documented the use of social support as positively influencing aspects of personal and family functioning. (71)

Social support provided by informal personal network members (extended family and friends) has been found to have the strongest relationship to any number of outcomes. Judge (71) suggests that there is a considerable positive impact on parent self-efficacy and personal control appraisals when professional help-giving practices mirror the features of informal support. When support from extended family and friends are not available, such caregivers may benefit from the formal social support provided by nurses, psychologists and other healthcare providers. (28) Therefore, healthcare professionals can help families of CWE to identify existing sources of informal and formal social support as well as hitherto untapped-but-potential sources of social support that matches family needs.

5.2 Strength of the Study

This study was unique as:

1. It was the first to evaluate caregiver burden of paediatric epilepsy among South African families and provides information (raw scores) and insight for use of this tool within similar groups.
2. It measured the impact of childhood epilepsy on the family utilizing a valid instrument which has been widely used in a variety of paediatric conditions.
3. An interviewer method was used, which has the advantage of maximizing response rate, and having few, if any missing item(s) as well as minimizing errors of misunderstanding.

4. Multivariate analysis allowed us to identify predictors of high impact from the associated factors.

5.3 Limitation of the Study

Although sample size calculation was not done, but I recruited as many caregivers of children with epilepsy as possible into the study. The sample size was limited by feasibility and time limitations. While the study might not have sufficient power to detect smaller effect sizes, it still allowed me to address my main research questions.

Our study population was limited to caregivers of CWE attending Charlotte Maxeke Academic Hospital epilepsy clinic, a tertiary care facility, rather than general population. Even though, we receive referral from all over Johannesburg and neighbouring provinces, this may limit generalizability of the results.

The investigator had to provide cut-offs without knowledge of scores for healthy control. Although PedsQL scale is a validated scale and is widely used in the literature in various settings. However, cut-offs defining groups who suffer high or low impact have not been generally described. From the theory of psychometric testing it is known that the actual values of scales are dependent on the population in which they are applied and hence cut-off values cannot easily be accepted from one population to another.(60) Previous analyses using the PedsQL scale used as cut-off values mean - 1SD (61) and mean - 2SD from a healthy control group (62) to define high impact. These values would not translate to the data from my study as the mean values were much higher (30 units) in these American studies.

In addition, the distribution of the PedsQL in my study was skewed and hence the PedsQL results were described using median and inter-quartile range rather than mean and standard

deviation. Therefore, I decided using the 25th and 75th percentiles to create impact groups; i.e. defining the lowest and highest 25% values. If I have had a symmetrical distribution using the mean and standard deviation, I would have used the mean – 1 SD and hence defined a high impact group with about 16% of the patients. As such my approach is slightly more clinically conservative, taking more participants into the high impact group.

CHAPTER SIX - CONCLUSIONS AND RECOMMENDATIONS

6.1 Conclusions

The following conclusions can be drawn from the current study;

1. This study provides raw scores of HRQOL and family functioning of caregivers of CWE with high impact of caregiver burden.
2. There is a strong positive correlation between caregivers' HRQOL and family functioning.
3. Multivariate linear regression analysis confirmed the caregiver's level of education and the child's seizure frequency as independent predictors of high impact of caregiver burden in paediatric epilepsy.
4. The families of CWE function better when they enjoy social support in an informal manner, like that from extended families and friends.

6.2 Recommendations

1. Caregivers and their families, especially those whose children have high seizure frequency, would benefit from added social support from extended families and friends to alleviate the constant caregiving demands.
2. Appropriate combination therapy and early monotherapy whenever possible, may help reduce the impact of paediatric epilepsy on both the child and parents.
3. Provision of epilepsy education for parents, siblings and extended families in the form of weekly talks, leaflets and pamphlets as well as counselling services will enhance the families' knowledge, strengthen their coping strategies and improve total functioning (reduce impact).

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APPENDIX A

INFORMATION SHEET FOR CONSENT

Study title: Impact of caregiver burden in paediatric epilepsy at Charlotte Maxeke Johannesburg Academic Hospital, Johannesburg.

Investigator: Dr. Umar A. Sabo

Supervisor: Prof. Gail Scher

Institution: Charlotte Maxeke Johannesburg Academic Hospital and the University of Witwatersrand

Good day. My name is Dr. Umar Sabo. I am currently working in the Division of Paediatric Neurology, Charlotte Maxeke Johannesburg Academic Hospital.

Introduction: We are conducting research on the burden of care of children with epilepsy and its impact on their family. Research is a process by which one learns and gains information. This study aims to learn more about the effect of burden of caring for children with epilepsy on the caregiver and assess its impact on their family functioning.

Invitation to participate: We are asking for your permission to participate in this study. It is important that you understand the voluntary nature of your participation in this study.

What is involved in the study: We are requesting you to answer questions to the best of your ability from the set of standardized questionnaires. We are also requesting for your permission to use your child medical records in the study.

Risk: There could be psychologic effect to you but anyone that shows signs of psychological distress from his/ her participation in the study will be offered psychotherapy.

Benefit: The research aims to determine the effects of caregiver burden in paediatric epilepsy on carers and its impact on their family functioning. Those caregivers and their family that are found to be functioning poorly (negatively impacted by the burden of care) will be referred to a psychologist, a social worker or epilepsy support group which at the moment is not part of our

routine care**.** The services may eventually improve their quality of lives which in turn will have a positive impact on the lives of their children.

Voluntary participation: Participation is entirely voluntary and declining will not affect your child's treatment in any way. If you want to withdraw your child from the study at any time, you are free to do so and it will not affect your child's treatment in anyway.

Confidentiality: Personal information will be treated as confidential. It may be disclosed only if requested by law. Note that absolute confidentiality cannot be guaranteed. Organizations that may inspect and/or copy your research records for quality assurance and data analysis include groups such as the Research Ethics Committee.

You and your child will remain anonymous even if the results are to be published.

Contact details of the researcher: for further enquiries, you can contact me through;

071 872 8300

Thank you.

INFORMED CONSENT

- I hereby confirm that I have been informed by the study doctor, Dr Sabo, about the nature, conduct, benefit and risks of the clinical study.
- I have also received, read and understood the above written information (Information sheet for Consent) regarding the clinical study.
- I am aware that the results of the study, including my personal details and my child's diagnosis will be anonymously processed into a study report.
- I may, at any stage, without prejudice, withdraw my consent and participation in the study.
- I have had sufficient opportunity to ask questions and of my own free will, declare myself prepared to participate in the study.

Caregiver

Printed Name Signature / Mark or Thumbprint Date and Time

I, Dr Umar A. Sabo, herewith confirm that the above participant has been fully informed about the nature, conduct, benefit and risks of the above study.

Researcher:

Printed Name Signature Date and Time

Translator / Other person explaining informed consent…… **(Designation):**

Printed Name	Signature	Date and Time

Witness:

Printed Name	Signature	Date and Time

APPENDIX B

DATA SHEET

CAREGIVER DATA (to be completed by the caregiver)

Study No: ……..

Phone No:……..

Age:……..

Gender: M ☐ F ☐

Religion: Christianity ☐ Hindu ☐ Islam ☐ Judaism ☐ Others ☐

Marital status: Married ☐ Divorced ☐ Widowed ☐ never married ☐

Separated ☐

Employment: Public service ☐ Private ☐ Trading ☐ artisans ☐

Retirees ☐ Unemployed ☐ others ☐

Education: Grade 7 ☐ Grade 10 ☐ Grade 12 ☐ college diploma ☐

Degree ☐

Caregiver's income/ month: < R2000 ☐ R2000 - 5000 ☐ R11000 – 30000 ☐

R31000 – 50000 ☐ > R50000 ☐

Family's income/ month: <R5000 ☐ R5000 – 20000 ☐ R21000 – 50000 ☐

R50000 – 100000 ☐ .R100000 ☐

Housing: House ☐ Flat ☐ RDP ☐ Shack ☐

Support: Spouse ☐ Extended family ☐ Friends ☐ NGO ☐ Govt grant ☐

Time spent on patient care: < 1hr ☐ 1-6hrs ☐ 7-12hrs ☐ 13-24hrs ☐

(hours per week) 24-48hrs ☐ 48-72hrs ☐ >72hrs ☐

APPENDIX B

DATA SHEET II

CLINICAL INFORMATION (to be completed by the researcher)

Study No:……..

Age of patient……

Age at onset of epilepsy……

Duration of epilepsy……

Average No of seizures per month in the past 3 months:

For complex partial: None ☐ 1-4 ☐ 5-10 ☐ > 10 ☐

For simple partial, myoclonic, absence: None ☐ 1- 20 ☐ > 20 ☐

For generalized tonic-clonic/ tonic/ atonic: None ☐ 1 ☐ 2-5 ☐ >5 ☐

No of AEDs taking by the patient: None ☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ >4 ☐

EEG finding(s): ……

CT/ MRI Brain finding (s):…….

Co-morbid condition(s): Cerebral palsy ☐ ADHD ☐ Depression ☐

Behavior problem ☐ Anxiety ☐ Others specify……..

APPENDIX C

PedsQL™

Family Impact Module

Version 2.0

PARENT REPORT

DIRECTIONS

Families of children sometimes have special concerns or difficulties because of the child's health. On the following page is a list of things that might be a problem for **you**. Please tell us **how much of a problem** each one has been for **you** during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month**, as a result of your child's health, how much of a problem have **you** had with…*

PHYSICAL FUNCTIONING ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I feel tired during the day	0	1	2	3	4
2. I feel tired when I wake up in the morning	0	1	2	3	4
3. I feel too tired to do the things I like to do	0	1	2	3	4
4. I get headaches	0	1	2	3	4
5. I feel physically weak	0	1	2	3	4
6. I feel sick to my stomach	0	1	2	3	4

EMOTIONAL FUNCTIONING ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I feel anxious	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I feel frustrated	0	1	2	3	4
5. I feel helpless or hopeless	0	1	2	3	4

SOCIAL FUNCTIONING ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I feel isolated from others	0	1	2	3	4
2. I have trouble getting support from others	0	1	2	3	4
3. It is hard to find time for social activities	0	1	2	3	4
4. I do not have enough energy for social activities	0	1	2	3	4

COGNITIVE FUNCTIONING ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. It is hard for me to keep my attention on things	0	1	2	3	4
2. It is hard for me to remember what people tell me	0	1	2	3	4
3. It is hard for me to remember what I just heard	0	1	2	3	4
4. It is hard for me to think quickly	0	1	2	3	4
5. I have trouble remembering what I was just thinking	0	1	2	3	4

COMMUNICATION ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I feel that others do not understand my family's situation	0	1	2	3	4
2. It is hard for me to talk about my child's health with others	0	1	2	3	4
3. It is hard for me to tell doctors and nurses how I feel	0	1	2	3	4

*In the past **ONE month**, as a result of your child's health, how much of a problem have **you** had with…*

WORRY ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I worry about whether or not my child's medical treatments are working	0	1	2	3	4
2. I worry about the side effects of my child's medications/medical treatments	0	1	2	3	4
3. I worry about how others will react to my child's condition	0	1	2	3	4
4. I worry about how my child's illness is affecting other family members	0	1	2	3	4
5. I worry about my child's future	0	1	2	3	4

DIRECTIONS

Below is a list of things that might be a problem for **your family**. Please tell us **how much of a problem** each one has been for **your family** during the **past ONE month.**

*In the past **ONE month**, as a result of your child's health, how much of a problem has **your family** had with…*

DAILY ACTIVITIES ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. Family activities taking more time and effort	0	1	2	3	4
2. Difficulty finding time to finish household tasks	0	1	2	3	4
3. Feeling too tired to finish household tasks	0	1	2	3	4

FAMILY RELATIONSHIPS ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. Lack of communication between family members	0	1	2	3	4
2. Conflicts between family members	0	1	2	3	4
3. Difficulty making decisions together as a family	0	1	2	3	4
4. Difficulty solving family problems together	0	1	2	3	4
5. Stress or tension between family members	0	1	2	3	4

Pediatric Quality of Life Inventory™ (PedsQL™)

The Parent report of the **PedsQL™** 2.0 Family impact Module is composed of 36 items comprising 8 dimensions.

DESCRIPTION OF THE FAMILY IMPACT MODULE: Dimensions	**Number of Items**	**Cluster of Items**	**Reversed Scoring**	**Direction of Dimensions**
Physical Functioning	6	1-6	1-6	Higher scores indicate better functioning.
Emotional Functioning	5	1-5	1-5	
Social Functioning	4	1-4	1-4	
Cognitive Functioning	5	1-5	1-5	
Communication	3	1-3	1-3	
Worry	5	1-5	1-5	
Daily Activities	3	1-3	1-3	
Family Relationships	5	1-5	1-5	

SCORING OF DIMENSIONS:

Item Scaling	5-point Likert scale from 0 (Never) to 4 (Almost always)
Weighting of Items	No
Extension of the Scoring Scale	Scores are transformed to a 0 to 100 scale.
Scoring Procedure	**Step 1: Transform Score** Items are reversed scored and linearly transformed to a 0-100 scale as follows: 0=100, 1=75, 2=50, 3=25, 4=0 **Step 2: Calculate Scores by Dimensions** □ If more than 50% of the items in the scale are missing, the scale scores should not be computed, □ Mean score = Sum of the items over the number of items answered. **Step 3: Total Scores** □ The Total Score is the sum of all 36 items divided by the number of items answered □ The Parent HRQL Summary Score (20 items) is computed as the sum of the items divided by the number of items answered in the Physical, Emotional, Social, and Cognitive Functioning scales. □ The Family Functioning Summary Score (8 items) is computed as the sum of the items divided by the number of items answered in the Daily Activities and family Relationships scales.

Interpretation and Analysis of Missing Data

If more than 50% of the items in the scale are missing, the Scale Scores should not be computed.

If 50% or more items are completed: Impute the mean of the completed items in a scale.

APPENDIX D

R14/49 Dr Umar Abba Sabo

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)

CLEARANCE CERTIFICATE NO. M150658

NAME: (Principal Investigator)	Dr Umar Abba Sabo
DEPARTMENT:	Paediatrics Charlotte Maxeke Johannesburg Academic Hospital Paediatrics Epilepsy Clinic
PROJECT TITLE:	Impact of Caregiver Burden in Paediatric Epilepsy Charlotte Maxeke Johannesburg Academic Hospital
DATE CONSIDERED:	26/06/2015
DECISION:	Approved unconditionally
CONDITIONS:	
SUPERVISOR:	Prof Gail Scher

APPROVED BY: _____

Professor P Cleaton-Jones, Chairperson, HREC (Medical)

DATE OF APPROVAL: 23/09/2015

This clearance certificate is valid for 5 years from date of approval. Extension may be applied for.

DECLARATION OF INVESTIGATORS

To be completed in duplicate and **ONE COPY** returned to the Secretary in Room 10004, 10th floor, Senate House, University.
I/we fully understand the conditions under which I am/we are authorized to carry out the above-mentioned research and I/we undertake to ensure compliance with these conditions. Should any departure be contemplated, from the research protocol as approved, I/we undertake to resubmit the application to the Committee. **I agree to submit a yearly progress report**.

_____ _____

Principal Investigator Signature Date

PLEASE QUOTE THE PROTOCOL NUMBER IN ALL ENQUIRIES

APPENDIX E

PLAGIARISM CHECK REPORT

Prof G. Scher-

Turnitincopy.docx

ORIGINALITY REPORT

25%	15%	22%	%
SIMILARITY INDEX	INTERNET SOURCES	PUBLICATIONS	STUDENT PAPERS

PRIMARY SOURCES

1	www.hqlo.com Internet Source	3%
2	ANGELA M. McNELIS. "Concerns and Needs of Children With Epilepsy and Their Parents", Clinical Nurse Specialist, 07/2007 Publication	2%
3	www.jcomjournal.com Internet Source	2%
4	bmcpsychology.biomedcentral.com Internet Source	1%
5	hqlo.biomedcentral.com Internet Source	1%
6	Shore, C.P.. "Maternal adaptation to a child's epilepsy", Epilepsy and Behavior, 200408 Publication	1%
7	www.qub.ac.uk Internet Source	1%
8	annals.org Internet Source	1%