

The impact of epilepsy on children and adult patients' lives: Development of a conceptual model from qualitative literature

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ABSTRACT

Aim: This study aimed to develop a conceptual model of the impact of partial onset or generalized epilepsy on children and adults in order to guide the identification of endpoints that capture patient perspectives in new treatment trials.

Methods: A systematic literature search was conducted in Embase and Medline to identify qualitative research reporting the impact of epilepsy on child and adult patients' lives. The search identified 20 publications describing 18 qualitative studies. Qualitative results were extracted from these publications into structured summary tables separately for impact on children and adults.

Results: Results tables were reviewed by two qualitative researchers who identified 23 concepts/areas of impact. Concepts were largely universal between child and adult studies, although concept content did vary between age-groups, for example child relationship concerns were focused on developing friendships and problematic family relationships. For adults the concerns were problematic relationships with spouse or partner and fulfilling the family roles. Concepts influenced directly by epilepsy were cognitive, physical and seizure effects, other concepts such as future hopes, burden and self-esteem were influenced more indirectly by impact on other concepts. The 23 concepts were linked to form a conceptual model of the impact of epilepsy for patients guided by qualitative results reported by studies.

Conclusion: The conceptual model suggests potential areas of patients' lives that may be enhanced by effective treatment and allows for concepts of concern to both children and adults to be identified and explored as potential endpoints in trials of new epilepsy treatments.

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1. Introduction

Epilepsy is a complex disorder which affects patients' psychological health, independence, emotional adjustment and employment.¹ Standard treatment for epilepsy is monotherapy with an anti-epileptic drug (AED) or adjunctive therapy if attempts at monotherapy with AEDs have not resulted in seizure freedom.² Various types of surgery offer additional treatment options for patients with medically intractable epilepsy.

The clinical efficacy of epilepsy treatments is commonly measured in terms of seizure freedom or significant reduction in seizure frequency, such as 50%.³ However, in recent years there has been growing recognition of the value of capturing wider impact of treatments on patients' health-related quality of life (HRQL) as a measure of treatment benefit, and of the need to place clinical trial

evidence of clinical efficacy into the context of meaningful improvement for patients.⁴ Patient-reported HRQL is an example of a patient-reported outcome (PRO) defined as "any report coming directly from patients, without interpretation by physicians or others, about how they function or feel in relation to a health condition and its therapy".⁵ PROs take the form of carefully designed questionnaires used to capture and quantify the patient experience of treatment and treatment impacts. The role of PROs has become increasingly important in clinical trials that evaluate the effectiveness of medicinal products, where they represent the voice of the patient.⁶ Treatment impacts on HRQL have become important to evaluate in general and are particularly important for chronic conditions where survival may not be the most relevant outcome of a treatment, intervention or therapy.

However, PROs are only useful if they capture patient perspectives and priorities on relevant elements of health. In order to be able to develop a PRO measurement strategy it is important to develop an understanding of the impact a condition has from the perspective of patients. This can be achieved through the use of qualitative research methods which allow patients' view-points to be elicited without patients being constrained to

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pre-determined topics which may or may not be important to them.⁷ As a result, qualitative methods allow patients to identify and prioritise factors that are important to them and provide insight into the meaning and experience of health, illness and treatment for patients, as well as the wider impact of health, illness and treatment on their lives. As part of qualitative analysis, conceptual models can be developed to summarise interconnected areas of impact of a condition or treatment for patients. In turn conceptual models can be used to identify endpoints that can be captured by PROs in the evaluation of new treatments in clinical trials.⁸

Conceptual models for the patient impact of epilepsy have previously been developed as part of qualitative studies with paediatric or adolescent samples.^{9–11} In order to inform a PRO measurement strategy for evaluating new AED treatments for children and adults with partial onset or generalized seizures we were interested in developing a broader conceptual model which explores important common areas of impact experienced by epilepsy patients from childhood, into and throughout adulthood. The relatively large body of existing qualitative literature in the epilepsy field offers the opportunity to synthesise evidence from a wide range of epilepsy samples through literature review, offering greater variety and a broader investigation of perspectives and impact than is usually possible in a single qualitative study.

2. Methods

2.1. Qualitative literature search

Structured Embase and Medline searches were conducted in August 2010 to identify published qualitative research investigating the impact of epilepsy on adult and paediatric patients' lives. Searches were constructed by combining epilepsy and seizure subject heading search terms (exploded to include content linked to sub-headings) with a number of qualitative research methodology terms (e.g. qualitative research, focus groups, nursing methodology research, patient interviews, purposive sample, phenomenology, grounded theory, content analysis, thematic analysis, qualitative study, semi-structured interview, qualitative interview, qualitative methods, qualitative analysis) and patient perspective/quality of life terms (personal experience, patient perspectives, patient journey, illness journey, patient views, life story, patient attitudes, attitude to health, quality of life, QoL, health-related quality of life, HRQL) using a combination of subject heading, text, adjacent and near adjacent search terms as well as searching conventions for identifying use of single and plural of terms. Searches were limited to 'human' and 'English language'. Full search terms are shown in [Appendix A](#).

Search results from each literature database were imported into Reference Manager[®] software and checked for duplicates prior to abstract review. Abstracts of publications identified by these searches were screened to identify primary qualitative research among child and/or adult epilepsy patients. Studies were excluded if they:

- did not include partial onset epilepsy patients (with or without generalized seizures) or generalized epilepsy patients
- focused on surgical treatment
- were not qualitative research
- were conducted outside of developed countries
- focused on epilepsy as a secondary condition (e.g. where epilepsy is part of a wider syndrome).

Full text versions of publications reporting qualitative studies conducted in child and adult samples were retrieved and reviewed.

2.2. Conceptual model development

Study design, sample characteristics and themes relating to the impact of epilepsy on patients' lives were extracted into structured tables by two experienced qualitative researchers (CK and AN) with an emphasis on summarising results in as much detail as possible. Impacts reported in direct quotes from patients were included as well as thematic qualitative analysis results reported in these publications. One table summarised qualitative studies with child or adolescent epilepsy samples and a second summarised qualitative studies for adult epilepsy samples.

The researchers met to systematically work through the detailed qualitative results from both age-groups together, discussing impacts and moderators of impacts reported by each study. During the process of identifying impacts/moderators, different colours were used to distinguish impacts/moderators identified from child/adolescent epilepsy samples from those identified from adult epilepsy samples. Researchers then further discussed impacts in order to group conceptually similar impacts into concepts. Similarities and differences between impacts identified from child/adolescent and adult samples were considered as part of this process, referring back to the detailed content of the structured tables for clarification. Finally, concepts were linked to form a single conceptual model, guided by information summarised in the structured tables.

3. Results

3.1. Literature search and review

Embase and Medline searches identified 167 publications after checking for duplicates. Of these, full text versions of 42 publications were retrieved for further review. In total 20 qualitative publications were identified that reported results from 18 studies conducted with child, adolescent and/or adult partial onset or generalized epilepsy samples. Four studies, reported in 5 publications, focused on epilepsy in children,^{10,12–15} 1 on epilepsy in children and adolescents,¹⁶ 1 on epilepsy in adolescents,⁹ 1 on epilepsy in adolescents and young adults,¹⁷ 1 on epilepsy in young adults¹⁸ and 10 studies, reported in 11 publications, focused on epilepsy in adults,^{18–28} although one of these included patients as young as 15 years old.²¹

Most studies were conducted in the UK ($n = 8$) or US ($n = 5$), although studies were also conducted in Canada ($n = 2$), Sweden ($n = 2$) and Australia ($n = 1$). Three of the studies focused on specific experiences of certain ethnic or gender groups of adults with epilepsy. These were African-American females,²² people of Pakistani origin living in the UK²³ and women of child-bearing age.²⁶ Further details of the qualitative studies are given in [Table 1](#).

3.2. Conceptual model

Review of the extracted qualitative results from the 18 studies identified 23 concepts/impacts of epilepsy on patients' lives. These concepts are shown in the conceptual model ([Fig. 1](#)) and described more fully in the following sections.

3.3. Burden

Concerns about the effect an individual's epilepsy has on those around them (or burden on others) were reported by studies with children, adolescents and adults. Children with epilepsy reported friends, parents and siblings worried about them, perhaps too much, and were involved in helping them and keeping them safe.¹⁰ Adolescents and young adults with epilepsy reported friends who were alarmed by the diagnosis or by witnessing a seizure.¹⁷ Other

Table 1
Qualitative studies conducted with child/adolescent/adult epilepsy samples.

Children/adolescents/adults	Authors/date	Country	Sample	Study aim(s)	Qualitative methodology and analysis
Children	Hightower et al., 2002	US	N=8 Aged 9–12 years 3 females, 5 males Ethnicity: 2 white, 6 black Seizure type: Absence with secondary generalized tonic/clonic (n=1) Absence (n=1) Primary generalized tonic/clonic (n=4) Simple partial seizures (n=1) Complex partial seizures with second-degree generalization (n=1)	To address the question 'what are the lived experiences of school-aged children with epilepsy?'	Individual interviews Thematic analysis
Children	McNelis et al., 2007	US	N=11 Aged 7–15 years 6 males, 5 females Length of illness from 18 months to 5 years Seizure type: Generalized tonic/clonic Absence Complex partial Simple partial	To explore concerns and needs of children with epilepsy and their parents	Focus groups Thematic analysis
Children	Moffat et al., 2009	UK	N=22 Aged 7 years 4 months–12 years 6 months 11 males, 11 females Age at diagnosis 1–9 years, mean 6 years 1 month Seizure type: Simple partial (n=6) Complex partial (n=5) Generalized tonic-clonic (n=7) Absence (n=7)	To investigate children's perceptions of the impact that epilepsy has on their QOL	Focus groups and individual interviews Grounded theory approach
Children	Ronen et al., 1999, 2001	Canada	N=29 Aged 6–10 years, 4 months Children with active epilepsy i.e. at least 2 unprovoked seizures with the past 24 months Seizure types: Simple/complex partial (n=10) Complex partial and generalized tonic-clonic (n=5) Generalized tonic-clonic (n=4) Typical/atypical absence (n=7) Absence and generalized tonic-clonic (n=2) Myoclonic and absence (n=1)	To identify the different components of HRQL in childhood epilepsy in order to create an HRQL disorder-specific measure	Focus groups Textual analysis
Children/adolescents	Elliott et al., 2005	Canada	N=49 Aged 7–18 years (7–12 years n=18, 13–18 years n=31), mean age at interview 13.66 years 24 males, 25 females Mean age at seizure onset 6.18 years, range 0–14 Medically refractory seizures (intractable epilepsy)	To further build on understanding of how children and adolescents with medically refractory epilepsy perceive the impact of epilepsy on their QOL	Individual interviews Three phases of analysis; open coding, axial coding and selective coding
Adolescents	McEwan et al., 2004	[7,0]UK	N=22 Aged 12 years 4 months–18 years 0 months 6 males, 16 females Age at diagnosis 1 month–13 years 6 months Seizure type: Simple partial (n=9) Complex partial (n=5) generalized (n=16) Myoclonic (n=1)	To describe the experience of having epilepsy in adolescence To contribute to the understanding of the perceived impact of epilepsy on QOL in adolescence To explore any changes in QOL issues as the adolescent progresses towards adulthood To develop a disease framework for understanding the impact of epilepsy in adolescence	Focus groups Thematic analysis

Table 1 (Continued)

Children/adolescents/adults	Authors/date	Country	Sample	Study aim(s)	Qualitative methodology and analysis
Adolescents/young adults	Wilde and Haslam, 1996	[7,0]UK	N = 24 Aged 13–25 years 15 females, 9 males 54% of subjects had seizures begin between the ages of 6–12 years Seizure type: Complex partial seizures with secondary generalization (n = 11) Complex partial seizures without generalization (n = 7) Primary generalized tonic–clonic seizures (n = 5) Simple partial seizures (n = 1)	[7,0]To explore the issues affecting young people with fairly significant epilepsy attending outpatient clinics	Individual interviews Thematic analysis using the interview guide as a framework
Young adults	Raty et al., 2007	Sweden	N = 95 Aged 18–27 years Recruited 5 years earlier from treating hospitals Uncomplicated epilepsy sample (without associated neurological impairment e.g. mental retardation or motor impairments) Epilepsy well-controlled in 61.7% of patients and in remission in 17.6% (n = 18) Seizure type Generalized tonic–clonic (37.1%) Complex partial seizures with generalization (18.6%)	To illuminate the impact of epilepsy on daily life in young adulthood, described by patients' emotions	Mailed survey containing open-ended questions on subjects' experiences of everyday life in relation to epilepsy Content analysis. Theoretical framework identified during analysis
Adults	Bishop and Allen, 2003	US	N = 24 Aged 18–80, most 30–60 60% female 15% diagnosed in last 3 years, 27% diagnosed 10–20 years ago, 40% diagnosed more than 20 years ago 92% taking medication for seizure control 60.5% reported experiencing seizures monthly Seizure type: Simple partial (11.6%) Complex partial (23.3%) Tonic–clonic (25.6%) Mixed (more than one type 39.5%)	To identify the QOL domains that are important to persons with epilepsy	Mailed survey containing open-ended questions on QOL Open coding. Themes identified by finding relationships between topics
Adults	Kilinc and Campbell, 2009	UK	N = 52 Aged 19–57 All diagnosed with epilepsy after the age of 18 Duration of epilepsy 11 months–30 years 12 well-controlled for 2–22 years All currently taking AEDs	To explore the experience of stigma for adults with epilepsy using a phenomenological approach	Individual interviews Phenomenological approach. Significant statements were grouped into themes
Adults	McCorry et al., 2009	UK	N = 47 Median age 38, range 15–68 18 males, 29 females Median time since diagnosis 4 years (range 9 months–35 years) Diverse in whether patients had experienced an AED drug change or AED dose change at a recent hospital visit 10 had no seizure in the 6 months prior to the hospital visit	To explore the issue of decision making from the patients' perspective	Individual interviews (telephone) Grounded theory approach
Adults	Paschal et al., 2005	US	N = 10 All African-American females with epilepsy 2 married, 2 divorced, 1 widowed and 5 never married 4 had at least 1 child under 18 living at home 5 had governmental health insurance (Medicaid and/or Medicare), 3 had private insurance and 2 were uninsured 3 had full-time jobs, 7 were unemployed (although 1 of these was a full-time student) Age at epilepsy onset ranged from 26 to 46 Seizures were uncontrolled in 6 participants	To explore access to health care, help-seeking behaviours and adherence to treatment among African-American females with epilepsy	Individual interviews (telephone and in-person) Grounded theory approach

Table 1 (Continued)

Children/adolescents/adults	Authors/date	Country	Sample	Study aim(s)	Qualitative methodology and analysis
Adults	Raty et al., 2009	Sweden	N=19 Aged (20–64) 12 females, 7 males Sample varied in Seizure frequency (0–50 in the last year) Living conditions and family situation (14 living with partner, 2 living with parent(s), 3 living alone, 12 had children, 1 was pregnant) Occupation Duration of epilepsy (1–49 years) Medication (various AED, some only 1, most taking 2 or 3) Seizure type Tonic/clonic only (n=5) General partial complex (n=1) Partial complex only (n=4) Partial complex and tonic/clonic (n=7) Absence (n=1) Unclear (n=1)	To highlight epilepsy patients' conceptions of epilepsy and emotions related to those conceptions	Individual interviews Phenomenological approach. Relationships between conceptions were analysed
Adults	Rhodes et al., 2008	UK	N=20 Aged 18+ All of Pakistani Muslim origin No learning disability	To explore attitudes towards epilepsy, others' attitudes, impact on patients' lives and perceptions of disability among Pakistani people with epilepsy	Individual interviews (conducted in Urdu, Punjabi or English) Analysis coding frame was developed based on common themes and subthemes
Adults	Ridsdale et al., 2003	UK	N=15 Newly diagnosed epilepsy patients who had received a nurse-led intervention as part of an RCT Age within range 17–83 Approx. 50% male/female	To describe and assess a nurse intervention from the patients' point of view	Individual interviews (conducted 6-months after baseline of RCT) Analysis approach unclear
Adults or their family members	Sample et al., 2006	US	N=41 participants Either patients or family members, representing 31 individuals with epilepsy 7 aged 21 or less, most aged 22–64 13 females, 18 males Most with onset of seizures in childhood (18) Nearly half (14) had experienced a seizure in the previous 30 days Most (19) were taking medication, 1 had a vagal nerve stimulator A good proportion (13) had other major comorbid conditions	To discuss individuals' experiences with accessing epilepsy-related services and health care and what life with epilepsy is like	Focus groups Analysis approach unclear
Adults	Thompson et al., 2008	UK	N=15 All women of child-bearing age diagnosed with epilepsy Mean age 32.6 years Sample diverse in parity, including women with No children (n=5) 1 child (n=3, one pregnant with 2nd) 2 children (n=6, one pregnant with 3rd) 3 children (n=1) Epilepsy diagnosis/seizure type Temporal lobe (n=4) Absences (n=4) Frontal lobe (n=1) Tonic clonic (n=2) Juvenile myoclonic (n=1) Unknown (n=3)	To undertake an in-depth exploration of women's experiences of epilepsy and the impact it had on their lives	Individual interviews Analysis approach unclear
Adults	Velissaris et al., 2007, 2009	Australia	N=90 Mean age 36 years 54 males, 36 females Patients within a month of 1st seizure Seizure type Focal (secondary generalized) 51% Simple partial and/or complex partial 18% Tonic-clonic (primary generalized) 20% Tonic-clonic (unclassified) 11%	To perform a broad-based, prospective and longitudinal assessment of psycho-social adjustment following a newly diagnosed seizure	Individual interviews including structured Austin first seizure impact interview (AFSII) Thematic analysis to code AFSII cognitive items

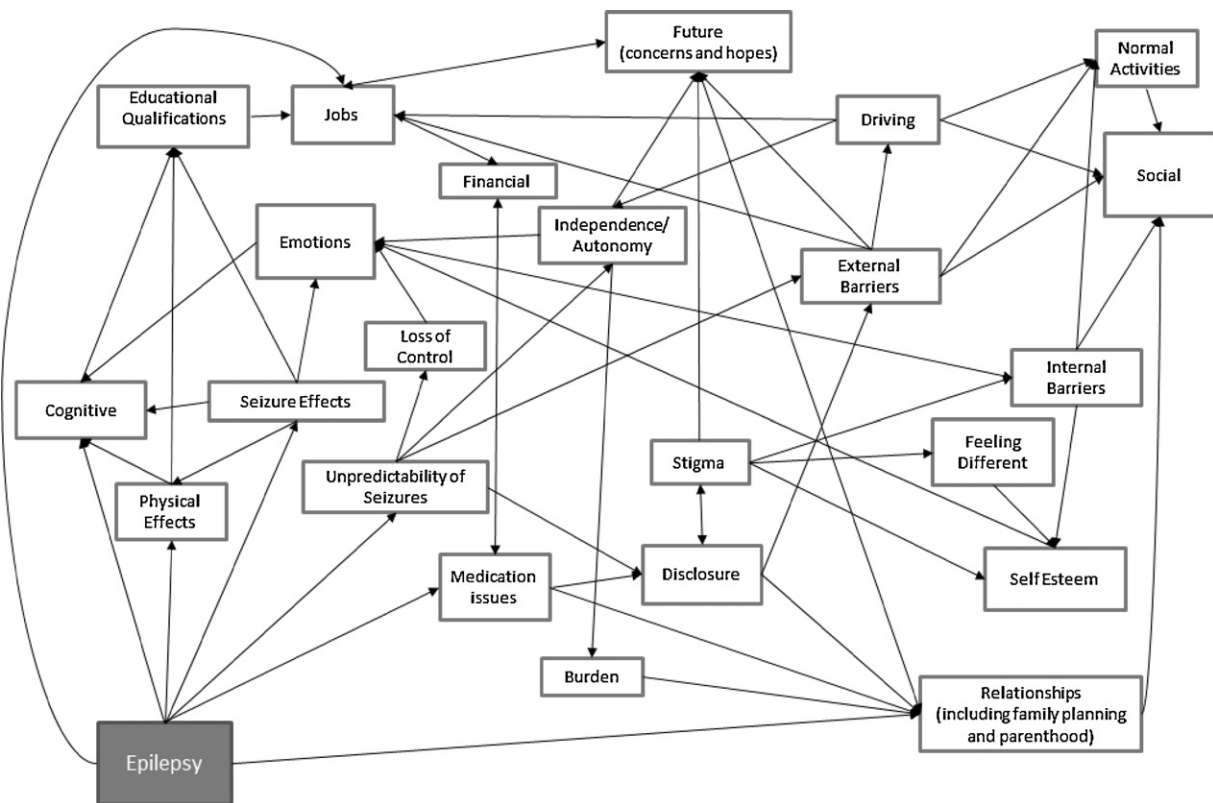


Fig. 1. Conceptual model of patient impact of epilepsy in children, adolescents and adults. This figure is the conceptual model of patient impact of epilepsy in children, adolescents and adults that was developed from the review of qualitative study results described in Section 3 of this paper. The model comprises 23 concepts/impacts of epilepsy on patients' lives that are described more fully in equivalent results sections in the paper. The 23 concepts/impacts of epilepsy were linked to form the conceptual model, guided by information reported by the published qualitative studies from which concepts were identified.

adult samples were concerned about the burden their epilepsy placed on others²⁵ and worried about the impact their epilepsy had on those around them.²⁰

3.4. Cognitive

The impact of epilepsy on cognitive functioning was widely reported by children, adolescents and adults. This included impaired attention/concentration,^{10,16,17,28} memory problems including forgetting things, short term memory loss, slower or more effortful retrieval of memories,^{9,10,19,28} perceived intellectual slow down or slower thinking speed,^{17,28} discontinuous or fragmented learning¹⁶ and muddled speech or difficulty finding words.²⁸ In a quantitative follow-up to a qualitative study with newly diagnosed adult epilepsy patients, emotional impact and pervasive loss of control resulted in greater cognitive complaints.²⁸

3.5. Disclosure

Whether or not to disclose having epilepsy was a significant issue for many patients.^{14,15,20,25} For children, disclosure to friends or school may be a necessary safety precaution.¹⁰ However, disclosure could have impact on job success^{17,29} and many patients chose not to disclose their epilepsy to mitigate social effects.²³ Disclosure was not always under the control of the individual, with patients expressing anxiety over disclosure by others or their epilepsy being revealed to others as a result of a seizure in public.^{9,10,29} Having to take epilepsy medications and not being able to do certain activities, e.g. drive or drink alcohol, marked patients out as different and so made it harder to conceal the diagnosis.^{9,17} Disclosure was also an issue for patients' families,

parents in particular may have strong views over whether their child's epilepsy should be kept secret.^{10,23}

3.6. Driving

From adolescence into adulthood, not being able to drive has significant impact for patients.^{9,17–19,24,25,27} Not being able to drive marks patients out as different¹⁷ and represents a loss of freedom¹⁹ as well as being a practical barrier to many normal activities.^{9,19,25} For many patients, failure to obtain a driving license or loss of a driving license on diagnosis of epilepsy was reported as the single most disabling aspect of having the condition²³ and a particularly hard impact for younger and middle-aged adults to cope with.²⁴

3.7. Educational qualifications

For children and adolescents, the cognitive effects of having epilepsy and its effect on school and exam attendance could impact educational attainment.^{9,10} For adults there was little motivation to attain educational qualifications if having epilepsy meant that no-one would employ them.²⁵

3.8. Emotions

The emotional impact of having epilepsy was universally reported by children, adolescents and adults. Types of emotional impact included anger,^{10,16–19} annoyance or frustration, particularly over limitations due to epilepsy;^{10,16,17,27} sadness, low mood, moodiness or depression;^{10,14–19,27} embarrassment,^{10,17,20,29} worry/anxiety;^{17,19} fear of seizures, of injury as a result of seizures, of being alone or doing things alone in case of seizures, of

embarrassment, of stigma or rejection,^{9,14,15,17,18,20,23,27,29} which for some adults could be significant enough to cause panic attacks.²⁰

3.9. External barriers

Restrictions and limitations to participating in normal activities were a key feature of the impact of epilepsy for patients. Some of these restrictions were imposed by law such as legal restrictions excluding epilepsy patients from driving or holding certain types of jobs.^{9,19,25} Others were imposed on patients by people in positions of authority on safety grounds, for example parents or teachers imposing social and activity restrictions as a protective measure,^{9,14,15,19,23} or not allowing children with epilepsy to leave home in adulthood.²⁵ These external barriers placed on epilepsy patients resulted in limited choices.¹⁹

3.10. Feeling different

Children, adolescents and adults with epilepsy reported feeling they were odd or different from those around them^{9,10,13,17,20,25} or that something was wrong with them.¹² This could be in terms of the way they see themselves and so expect others to see them^{14,15,20} or because they felt marked out as different because of having to take epilepsy medications, not driving or not drinking alcohol.^{14,15,17}

3.11. Financial

Adult patients spoke of loss of income following diagnosis with epilepsy because they were unable to return to work or to usual duties.²⁷ Affording epilepsy medications was also an issue for adult patients.^{22,25}

3.12. Future

Epilepsy patients of all age-groups had both concerns and hopes for the future. For children and adolescents, concerns focused on not being able to participate in usual social activities, manage a seizure in social situations, drive, leave home, live alone, form romantic relationships, get jobs, keep safe or have children.^{9,10,17} Adult patients' concern for the future focused particularly on pregnancy and parenthood.¹⁸ Children and adolescents hoped they might grow out of epilepsy,^{9,10} adolescents and adults hoped for a normal epilepsy- or seizure-free life in future.^{17,18} Adult patients tried to hold on to hope through a continuing search for new and more effective medications and interventions.²⁵

3.13. Independence/autonomy

Independence and development of autonomy were issues for children and adolescents with epilepsy.^{9,14,15} For adult patients, loss of freedom and independence was an important impact of epilepsy.^{19,23,25} Having epilepsy meant they had to change plans and in some cases it resulted in loss of their life plan.^{19,25} For some it meant they were unable to live independently²⁵ and in general having epilepsy limited their choices or options^{19,23} and their ability to do the things that they wanted to do.¹⁹

3.14. Internal barriers

In addition to legal restrictions or those imposed by others, many epilepsy patients restricted their own activities through fear of injury,^{9,17} fear of embarrassment as a result of seizure,^{9,29} and fear of rejection or stigma as a result of disclosure.^{9,17,23} Adult patients' fear of being or doing things alone or fear of having a

seizure in public could be particularly limiting in terms of usual activities they were able to engage in.^{18,20}

3.15. Jobs

Adolescents and adult patients reported difficulties getting a job because of epilepsy and had experienced rejection by employers.^{17,18,23,25} Patients with jobs found them hard to keep²⁵ and younger and middle-aged patients found job loss particularly hard to cope with.²⁴ Some patients reported restrictions in terms of types of work, e.g. seizures caused by sitting in front of a computer, types of career and levels of responsibility.^{18,19,29} Others were simply unable to work due to the severity of their condition.^{19,25} The impact of epilepsy on job restrictions and ability to work resulted in loss of career goals.²⁵

3.16. Loss of control

Feelings of loss of control were reported by children, adolescents and adult patients.^{16,27} This was experienced in relation to fear of seizure recurrence,^{10,19,27} control over their body and disruption to goals and plans.²⁷

3.17. Medication issues

Some medication issues related to the hassle of taking medications, for example remembering/forgetting to take medications, medications making it hard to conceal having the condition.^{9,10,24} Other issues related to the side effects experienced from medications, including stomach pains, light sensitivity, tiredness, metabolism, energy levels, memory problems and rashes.^{9,10,12,14,15,18,19,25,27} Patients were concerned about the safety of medications and raised concerns about addiction,¹² uncertain effects and interactions with other drugs.^{18,26} Women of child-bearing age had particular concerns about the interaction of epilepsy medications with contraceptives and their effects on a developing foetus.²⁶ These women had to make difficult treatment choices in order to balance risks against safety for seizure, contraception, avoiding unwanted pregnancy, healthy development of a foetus, and pain management during labour.²⁶

3.18. Normal activities

Epilepsy patients, particularly children, adolescents and young adults, reported restrictions to engaging in academic activities, social activities such as going out at night, drinking alcohol or going to night clubs¹⁶ and physical activities such as riding a bicycle or swimming.^{12,17}

3.19. Physical effects

Patients reported certain long-term physical effects of epilepsy that were experienced independently of seizures. Both child and adult patients reported experiencing fatigue and the need for more sleep.^{16,19,27} Children also experienced anergia/inertia.¹⁶ Other short-term physical effects were experienced intermittently during/after a seizure and are described in the seizure effects section below.

3.20. Relationships

Children with epilepsy experienced problems in relationships with their peers, parents and siblings.^{14,15} Adolescents and young adults reported difficulties developing romantic relationships, experienced rejection in relationships and felt frustrated by parental concern.^{9,17} Adult patients also reported problematic

relationships with their spouse or family members as a result of their epilepsy,^{17–19,22} in some cases with misunderstanding of or burden from epilepsy contributing to relationship breakdown.^{22,25,29} Epilepsy was seen to reduce the likelihood of marriage and patients experienced difficulties having children (conception, pregnancy and birth), with parenthood and fulfilling their family role.^{26,29}

3.21. Seizure effects

The experience of seizures had a number of effects for patients. Children and adolescents reported developing increased awareness of triggers of seizures and signs or auras that might signal a seizure was about to occur.^{9,10,12} In adults this could lead to hypervigilance to the signs of seizure.²⁷ Children reported engaging in preventative or precautionary behaviour such as temper control, drinking fluids, praying, keeping a regular bed time, exercise, good diet, resting well, using cold packs on their face, taking medications and informing others what to do in case of seizure.^{10,12} Adults also reported avoiding situations perceived as high risk.²⁷ During seizures patients could experience injury, incontinence, fear and other emotional distress^{10,12,16} and after seizures intermittent memory deficits, tiredness and headaches.^{10,16,27}

3.22. Self esteem

Epilepsy impacted patients' self-esteem through stigma and patients' feeling and being treated differently. Adolescent and adult patients reported feeling shame,^{17,18,29} guilt over the burden they placed on those around them, for scaring or alarming others or for ruining social occasions,^{18,29} self-doubt over being normal¹⁸ and feeling flawed or somehow lesser than others.²⁷

3.23. Social

Many patients experienced loneliness and were socially isolated.^{9,16,19,25} This was as a result of developing few friendships, losing friends and having a very limited social network beyond immediate family.^{9,17,19,23,25} Some patients found it hard to get out and come into contact with others,²⁹ others avoided or were passive in social situations to avoid drawing attention to themselves.²⁰

3.24. Stigma

Experienced, perceived and feared stigma as a result of epilepsy had significant impact for patients. Many children and adolescents felt others thought they were weird¹⁰ and experienced teasing, being talked about, being laughed at and bullying.^{9,10,14,15,17} Children and adolescents were aware they were treated differently to siblings and peers by parents, teachers and peers and experienced overprotectiveness, activity restrictions and lack of privacy as a result.^{9,10} Adolescents and adults experienced prejudice, discrimination, being stereotyped and being misunderstood.^{17,19,25} They also experienced being treated differently, for example people being overprotective, or parents not allowing them to leave home.²⁵

3.25. Unpredictability of seizures

The unpredictability of seizures was a key feature of epilepsy that explained many of the wider impacts for patients.^{16,18,20,23}

3.26. Moderators of impact of epilepsy

It should be acknowledged that the negative impacts of epilepsy summarised by the conceptual model and reported in the above

sections are not experienced by all patients or may only have been experienced soon after seizures were first experienced. A number of the qualitative publications identified factors that moderated the impact of epilepsy for patients and ways that patients had found to reduce the impact of epilepsy. These included access to medical support and the quality of medical support^{10,17,18,25} and support from close friends, family and others.^{12,19,22} Increased knowledge of epilepsy by patients and those around them helped, as well as learning to live with seizures or other aspects of the condition.^{9,12,14,15,18,20,22} Acceptance of the condition by themselves and others was also important.^{10,17,28} Some patients developed resilience, coping strategies and strategies for taking or restoring control.^{10,16,27} Finally some adult patients found music, spirituality and volunteering helped.²⁵

4. Discussion

This study reviewed results from 18 qualitative studies conducted with child, adolescent and adult patients with epilepsy to develop a conceptual model of the impact of partial onset or generalized epilepsy on children and adults. The resulting complex model proposes wide-ranging impact on 23 concepts and inter-relationships between areas of impact.

Health status outcomes can be categorised to identify a proximal-distal continuum of health status outcomes with clinical data e.g. signs and symptoms, representing the most direct indication of the presence of disease. As outcomes move increasingly further away from the indication of disease they become increasingly global and represent more distal outcomes e.g. from disease-specific functioning to general functioning and general well-being outcomes.³⁰ The conceptual model enables those researching epilepsy treatments or working with epilepsy in clinical practice to consider a broad range of potential outcomes of importance to epilepsy patients that could be influenced through treatment or other intervention. The structure of the model differentiates proximal concepts, influenced more directly by the condition (and located closer to 'Epilepsy' in the model) from more distal concepts, influenced indirectly by impact on other concepts. This offers useful guidance to those planning clinical trials who are limited in the number of endpoints that can be incorporated for reasons of statistical robustness, as concepts in the model that are proximal health status outcomes identified by patients e.g. Physical Effects, Cognitive, Seizure Effects, are those that are most likely to be directly changeable through AEDs, and therefore of value to incorporate into a clinical trial endpoint model.

The conceptual model also indicates aspects of patients' lives that could possibly be influenced through successful AED treatment that would be of value to monitor in large observational late phase studies of epilepsy patients where the focus is on longitudinal data in real-world settings. For example an AED treatment that improves aspects of the patients' lives associated with the concept of Physical Effects may lead to a long term benefit in terms of Educational Qualifications. Looking beyond clinical studies, the model also presents useful information to those working in clinical practice who have a focus on management of epilepsy patients as it indicates aspects of the patients' lives of concern to patients that are unlikely to be improved with current AED treatments because of their distal position in the model e.g. Self-Esteem, Stigma, Internal Barriers. These may warrant an alternative interventional approach and indicate related aspects of patients' lives that such targeted interventions might positively influence, for example targeting issues of Stigma may improve patients feelings towards the future, and ease difficulties around patient disclosure of epilepsy therefore improving relationship issues.

At the start of the data extraction and literature review process, the level of similarity of impact experienced by children and adults

with epilepsy was unknown. Previous conceptual models developed in this area have focused on quite specific age-groups/developmental stages.^{9,10} However, it is striking how many of the concepts were common to child/adolescent and adult patient samples. Only three of the concepts, 'driving', 'jobs' and 'financial' were not identified from studies with children as well as adults. Of these, impacts on 'driving' and 'jobs' were experienced from adolescence and studies conducted with children with epilepsy listed not being able to drive or get jobs as a concern for the future. Impacts on 'self-esteem' were also reported mainly by adults, however, this concept was closely related to the concept of 'feeling different', an impact more widely, but not uniquely reported by studies conducted with children with epilepsy. It may be that for children, impacts on 'self-esteem' are encompassed within 'feeling different'. The number of concepts common across age-groups supports the validity of developing the broad conceptual model covering impact for children and adults with partial onset or generalized epilepsy and of using the model to guide a PRO measurement strategy for the evaluation of new treatments for these groups by identifying important endpoints commonly impacted from childhood into adulthood.

While most concepts were common to children/adolescents and adults, in several cases the content of a concept varied across age-groups and this would need to be reflected in PRO instrument content. For example, within the concept of 'relationships', impacts for children were focused on friendships and relationships with parents/siblings, whereas for adults the focus was on relationships with their spouse or partner and fulfilling family roles. Again, concerns for the future among children and adolescents with epilepsy included difficulty forming romantic relationships and potential problems having children. However, a PRO instrument asking about experiences of this type of impact would be irrelevant to children with epilepsy. Equally impact on 'independence/autonomy' for children and adolescents with epilepsy focused on development of independence and autonomy, whereas for patients diagnosed in adulthood the impact was in terms of loss of freedom and limitation of options. Along with more obvious requirements for instruments e.g. related to children's reading ability, this means that for many concepts, it is unlikely that the same PRO instruments could be used with children and adult patients with epilepsy.

Notable by its absence from the conceptual model is any concept of 'seizure frequency', the most common clinical endpoint evaluated in treatment trials. This absence suggests that frequency of seizures per se may not be an important aspect of seizures for patients. Instead, unpredictability of seizures appeared key to a number of wider HRQL impacts for children and adults with epilepsy. Also patients' hopes for the future focused on being epilepsy or seizure free rather than for a reduction in seizure frequency. This is in line with quantitative research which found that in a clinical trial of AED treatment for adults with refractory complex partial seizures, only patients who became seizure free for the final 12 weeks of a 28-week trial follow-up reported significant HRQL improvements.³ This is a relatively long follow-up period by usual AED clinical trial standards. Authors of two other publications reporting AED trial results have noted changes in HRQL were not proportional to seizure control³¹ and commented that 3–4 months, a common timescale for AED efficacy trials, is probably too short to expect much change in HRQL.³² The conceptual model would suggest this may be because while AED treatments may have achieved seizure reduction, they may not have reduced the unpredictability of seizures for patients. The time-scale issue is also interesting from the patient perspective. How long must a patient be free of seizures for them to perceive themselves as seizure-free and experience associated HRQL benefit? Consideration of where a patient is on their epilepsy trajectory could also be relevant, for example, for a new onset patient or one with seizures in remissions, seizure freedom may well be the only desirable outcome; while for those with uncontrolled epilepsy a

small reduction in seizure frequency may sometimes be helpful. Investigation of patient perspectives on seizure frequency and its relationship with HRQL may be an interesting focus for future qualitative and quantitative research. However, it is important to be cautious in interpreting the lack of appearance of 'seizure frequency' in the model as this could simply be a limitation in study report. The limitation of lack of access to original data sets where seizure frequency may have been discussed is discussed further below.

By reviewing results from a number of qualitative studies conducted with various epilepsy samples, development of this conceptual model could take into account many more patient perspectives than would usually be afforded by patients in a single qualitative study. It also serves the purpose of drawing together existing evidence to better inform any future qualitative work with a view to ensuring that additional studies add value to the body of literature. However, this approach does suffer from the significant limitation of lack of access to the original qualitative data-sets from these studies. As a result, development of the conceptual model in this study relied on and was limited by the level of detail of qualitative analysis results reported in peer-reviewed publications. This was further compounded by the fact that the aims of many of these studies were quite specific and dictated the focus of questions used during data collection as well as priorities for qualitative analysis and report of the results. For this reason, content of direct patient quotes reported in publications were checked for report of impacts as well as the higher level qualitative analysis results. As a result of this limitation, the conceptual model presented here should be considered exploratory.

A further limitation of the literature review approach taken here is that it was restricted to English Language articles and epilepsy patients who were capable participating in qualitative studies. Although the review clearly still included a wide range of samples and studies, these restrictions may have resulted in lack of representation of important patient perspectives in the model e.g. culturally different impacts and impacts for patients with limited ability to communicate.

Despite these limitations, the model offers a broad and interesting picture of impacts of epilepsy for patients and opens up a number of avenues for future research. The validity of the model can be evaluated in future qualitative work with epilepsy patients of different ages, which can also investigate interesting questions raised by the model, for example the relative importance of the concepts included in the model for patients, the relationship between seizure frequency and unpredictability of seizures, and patient definitions of what it means to be seizure free. The model also suggests areas of patients' lives that may be impacted by effective treatment for measurement with PRO instruments in clinical trials or for future focus of PRO instrument development.

5. Conclusions

The conceptual model developed from qualitative literature suggests potential areas of patients' lives that may be enhanced if epilepsy symptoms were improved and allows for concepts of concern to both children and adults to be identified and explored as potential endpoints in trials of new epilepsy treatments.

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Appendix A. Appendix A

See Tables 1A and 2A.

Table 1A

Embase search 1996–2010 conducted 10th August 2010.

Search	Results	
1	exp EPILEPSY/	73030
2	exp SEIZURE/	43290
3	1 or 2	99966
4	qualitative research/	10293
5	focus group\$.mp.	13497
6	nursing methodology research/	10506
7	(patient\$ adj4 interview\$).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer]	23147
8	purposive sample/	76
9	INTERVIEW/	68495
10	phenomenology/	4700
11	personal experience/	4175
12	grounded theory/	573
13	content analysis/	1395
14	thematic analysis/	714
15	(patient\$ adj2 perspective\$).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer]	3997
16	(patient\$ adj2 journey\$).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer]	292
17	illness journey\$.mp.	7
18	(patient\$ adj2 view\$).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer]	3126
19	life stor\$.mp.	440
20	qualitative stud\$.mp.	9759
21	semi structured interview/	4109
22	qualitative interview\$.mp.	2200
23	qualitative method\$.mp.	3218
24	qualitative analysis/	22499
25	"quality of life"/	133429
26	QoL.mp.	14795
27	HRQL.mp.	1896
28	health related quality of life.mp.	15142
29	patient attitude/	28894
30	attitude to health/	42292
31	11 or 15 or 16 or 17 or 18 or 19 or 25 or 26 or 27 or 28 or 29 or 30	209626
32	4 or 5 or 6 or 7 or 8 or 9 or 10 or 12 or 13 or 14 or 20 or 21 or 22 or 23 or 24	131043
33	3 and 31 and 32	177
34	limit 33 to (human and English language)	138

Table 2A

Medline search 1996–2010 conducted 10th August 2010.

Searches	Results	
1	exp Epilepsy/	49292
2	exp Seizures/	14408
3	1 or 2	49433
4	Qualitative Research/	8660
5	Focus Groups/	9778
6	Nursing Methodology Research/	12134
7	(patient\$ adj4 interview\$).mp. [mp=title, original title, abstract, name of substance word, subject heading word, unique identifier]	9282
8	purposive sample\$.mp.	916
9	interviews as Topic/	23169
10	phenomenological research.mp.	156
11	personal experience\$.mp.	4096
12	grounded theory.mp.	2986
13	content analysis.mp.	5099
14	thematic analysis.mp.	1369
15	(patient\$ adj2 perspective\$).mp. [mp=title, original title, abstract, name of substance word, subject heading word, unique identifier]	3118

Table 2A (Continued)

Searches	Results	
16	(patient\$ adj2 journey\$).mp. [mp=title, original title, abstract, name of substance word, subject heading word, unique identifier]	194
17	illness journey\$.mp.	7
18	(patient\$ adj2 view\$).mp. [mp=title, original title, abstract, name of substance word, subject heading word, unique identifier]	2350
19	life stor\$.mp.	343
20	qualitative stud\$.mp.	8015
21	semi structured interview\$.mp.	5555
22	qualitative interview\$.mp.	1755
23	qualitative method\$.mp.	2525
24	qualitative analysis.mp.	3464
25	"Quality of Life"/	68949
26	QoL.mp.	10506
27	HRQL.mp.	1505
28	health related quality of life.mp.	11937
29	Attitude to Health/	40768
30	11 or 15 or 16 or 17 or 18 or 19 or 25 or 26 or 27 or 28 or 29	117002
31	4 or 5 or 6 or 7 or 8 or 9 or 10 or 12 or 13 or 14 or 20 or 21 or 22 or 23 or 24	68549
32	3 and 30 and 31	88
33	limit 32 to (English language and humans)	85

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