



Support values through the eyes of the patient An exploratory study into long-term support of persons with refractory epilepsy

Wil H.E. Buntinx^{a,*}, In Yu Tan^b, Albert P. Aldenkamp^{b,c}

^a Governor Kremers Center, Maastricht University Medical Center, Maastricht, The Netherlands

^b Epilepsy Center Kempenhaeghe, Heeze, The Netherlands

^c Maastricht University Medical Center, Maastricht, The Netherlands

ARTICLE INFO

Article history:

Received 3 November 2017

Revised 5 January 2018

Accepted 11 February 2018

Available online xxxx

Keywords:

Person-centered care

Long-term support

Refractory epilepsy

Quality of care

Patient values

Person-centeredness

ABSTRACT

A significant group of persons affected by refractory epilepsy require a wide range of long-term support from professionals as well as informal support givers. To enhance person-centeredness, it is important to know which patient values are associated with long-term support.

An Internet survey produced a total of 1176 statements from 289 persons with epilepsy for analysis. Statements were expressed in the respondent's own words and were related to positive experiences (704) as well as possible areas of improvement (472) regarding the support received in the past six months. Thematic summaries of the survey results were presented to four focus groups of persons with epilepsy and proxies with a request to formulate the most important patient values which should be observed in long-term support. In iterative sessions with a panel of epilepsy experts, a framework of twelve value domains was formulated, organized in four major value clusters.

An example of how these value domains can be operationalized for use in practice is next presented as a checklist to help evaluate the support patients with long-term support needs actually receive. Correspondingly, two sets of reflective questions were formulated for informal and professional support givers for the purpose of evaluating and improving person-centeredness in their support practices.

© 2018 Elsevier Inc. All rights reserved.

1. Introduction

In the Netherlands, about 120,000 individuals have been diagnosed with epilepsy. After appropriate diagnosis and treatment, the majority of these subjects are well-controlled by medication, and their outlook is good [1]. There are, however, about 36,000 cases, which are socially affected by refractory epilepsy and in need of long-term support in order to enhance their functioning and quality of life [2,3]. Kalsbeek and Plateel did carry out a Dutch study about quality criteria from the patient's perspective in medical diagnosis and early treatment of epilepsy [2], but it is still not clear what patients perceive as being important in long-term care and support situations that extend beyond early diagnosis and treatment.

Literature on this topic is scarce. To ascertain patients' views on how epilepsy healthcare services should be organized, Elwyn et al. [4] held focus group interviews, but the number of patients was low – 19 individuals in 5 groups – and persons with learning disability were excluded. Miller et al. [5] studied the needs of persons with epilepsy but focused exclusively on the elderly and on self-management. Wagner et al. [6] undertook surveys to investigate patient needs with a special

interest in support by community partners. In the extensive review by Mahendran et al. [7] on unmet healthcare needs in patients with epilepsy, the most recent paper on this subject was published in 2001.

We, therefore, undertook an exploratory study to ascertain patients' views on support values.

We use “support” as a collective term for ‘resources and strategies that aim to promote the development, education, interests, and personal well-being of a person and enhance individual functioning’ [8,9]. Supports to enhance individual functioning may stem from professional (for example: neurologists, nurses, social workers, psychologists, teachers, and coaches) as well as from informal (such as parents, partners, children, relatives, colleagues, and volunteers) resources. Further, there are multiple forms and functions of supports ranging from monitoring, coaching, instructing, offering personal assistance, facilitation, or the use of technical aids. In the present study, the concept of “value” is defined as a person's subjective appreciation and esteem with respect to support received; it is not used to monitor healthcare outcome relative to costs as, for example, defined by Michael Porter [10].

The present study was initiated and sponsored by two Dutch advocacy associations: the Dutch Epilepsy Association¹ and the Family and

* Corresponding author.

E-mail address: wil.buntinx@gmail.com (W.H.E. Buntinx).

¹ Epilepsievereniging Nederland (EVN), <http://www.epilepsievereniging.nl>.

Parents Association for Intensive Support in Epilepsy.² The project was supported by the care departments of SEIN and Kempenhaeghe, two Dutch tertiary referral centers for epilepsy. The purpose of the project was to explore support values from the person's perspective to validate future actions such as:

- promoting a person-centered focus in the work of neurologists, nurses, direct support staff, psychologists, and social workers in the field of long-term epilepsy support;
- empowering persons with epilepsy as well as their advocacy groups by raising awareness of legitimate expectations with respect to support; and
- facilitating development of quality-of-care instruments such as assessment scales, checklists, or guidelines.

The research questions to be answered in this study were as follows:

1. What do persons with epilepsy, who are receiving long-term support, report about what they consider to be “good” and what they consider to be “areas of improvement” in the support received in the past 6 months?
2. On the basis of the answer to research question 1: what do focus groups of persons with epilepsy and their proxies formulate as a framework of values to be observed when delivering person-centered support in long-term epilepsy?
3. How can answers to research questions 1 and 2 be converted into reflective questions for patients, informal support givers, and professional support givers in practice?

A qualitative research path was followed to reach an interpretative understanding of experiences of persons with epilepsy regarding long-term support (Ritchie et al. [11]). A central feature of the procedure was to establish a dialogue between the researchers and persons with epilepsy who are involved in receiving or offering long-term support. This dialogue was started with a survey submitted to the membership of the initiators and continued in iterative sessions with focus groups of those concerned and a discussion panel of stakeholders to arrive at consensus on a framework of values [12,13].

This research was approved by the boards of both participating patient organizations: the Epilepsy Association and the Family and Parents Association for Intensive Support in Epilepsy. All participants were informed about the purpose of the research project and were completely free to complete the questionnaire. Questionnaires were submitted anonymously.

2. Method

The first research question was addressed by a survey aimed at sampling personal experiences of good practice support as well as aspects of support that require improvement from the perspective of the person with epilepsy. The second question was addressed through focus groups; they reflected on the results of the survey and were invited to reach consensus on important characteristics of “good support”. A discussion panel of experts acted as a sounding board to comment on the outcomes of both the survey and the focus group studies to enhance consensus on the final value framework. This panel consisted of two neurologists and five self-advocates, as well as a psychologist and a social worker, both of whom were working in residential services for persons with epilepsy. The third question was answered using an iterative consensus procedure carried out with the discussion panel, resulting in a checklist of reflective questions for patients, informal support givers, and professionals to enhance person-centered practices.

The Internet survey was conducted among members of the Dutch Epilepsy Association and of the Family and Parents Association for Intensive

Support in Epilepsy. An invitation to take the survey was posted on their websites as well as those of the epilepsy centers and also distributed to the e-mail addresses of members of the Dutch Epilepsy Association. Following this approach, the potential (maximum) number of respondents which could be reached was 2100. The questionnaire used was edited in two formats: (1) for the person with epilepsy and (2) for a member of the social network of a person with epilepsy who acted as proxy. The latter concerned persons with epilepsy, who even with help, were unable to complete the questionnaire (e.g., young children, persons with intellectual disability or other severe communication impairment).

Samples of “good support” as well as of “areas of improvement” were collected using two open questions. These questions addressed support in general (1. ‘What do you appreciate about the support as currently received?’ – 2. ‘In what way could your present support be improved?’). The same two open questions were asked with respect to each of a number of specific quality-of-care domains that were presented to the respondents (Table 1, domains 11–18). This was done to broaden the respondent's scope of perception and to avoid too narrow a perspective, which could result in reporting only about a particular or recent experience of the respondent.

In order to organize the resulting experiences, a content analysis was performed on all answers to the first question of what was seen as “good” support and next on all answers to the second questions of what was experienced as “areas of improvement”. Therefore, every answer was coded according to the “quality of support domains” (Table 1), developed by the Dutch association of service providers in disability care [15]. After organizing the answers to the open questions, they were used as input for drafting brief thematic descriptions for each support category. These descriptions focused on identifying specific valued topics that were present in either “good support” or “area of improvement” statements. These topics were presented as vignettes to the focus groups. Vignettes contained the most frequently reported subjects in each support domain.

Four focus groups were held, each lasting 2 h, to discuss the vignettes and to identify and formulate key values with respect to the quality of long-term support. Members of the focus groups were persons who indicated in the survey that they were willing to participate in such groups. They were further selected to represent the following persons:

- adults (>18) with epilepsy (n = 6);
- parents of children (<18) with epilepsy but no intellectual disability (n = 6);
- parents of children (<18) with epilepsy and intellectual disability (n = 4); and
- proxies of adults (>18) with epilepsy and intellectual disability (n = 5).

The output from the focus groups was then integrated by the first author to construct a draft framework of values in long-term support and to formulate a set of reflective questions to operationalize the values. This process was evaluated and commented on in four iterations by the discussion panel of experts that approved the final edition of the framework and reflective questions. The resulting set of questions is an exemplary translation of value framework that can be used, for example, in educational settings or team evaluation in service settings to raise awareness about person-centered support.

3. Results

3.1. Subjects

A total of 289 persons completed the Internet survey: 114 persons with epilepsy and 175 proxy respondents (parents, siblings, significant others), equivalent to 14% of those invited to participate. Respondent characteristics are shown in Table 2.

² Stichting Zorgintensief & Epilepsie (ZIE), <http://stichtingzie.nl>.

Table 1

Coding categories for answers to open questions about what was considered “good” and “areas of improvement” in relation to long-term support being received by persons with epilepsy and their proxies.

Code	Support domain ^a	Respondent's statement relating to...
<i>Quality of life – content-related</i>		
11	Physical well-being	Care related to health and physical condition; body hygiene; prevention of health problems
12	Emotional well-being	Enhancing mental health; general life satisfaction; avoiding stress
13	Interpersonal relations	Maintaining positive relationships with family, friends; communicating; intimate relationships
14	Community participation	Participation in school, work, leisure activities; moving around and using transportation; shopping; club activities
15	Personal development	Learning new skills; participating in a course or training that teaches new skills; developing one's talents
16	Material well-being	Home comfort; budget management; management of assets
17	Self-determination	Making choices; focusing on goals; planning; life direction
18	Rights	Self-advocacy; privacy; handling a complaint; participating in clients' council or patients' association
<i>Facilitating conditions</i>		
21	Competence of support givers	Expertise with respect to epilepsy
22	Collaboration between professionals	Collaboration and exchange of information between professionals within a direct support team as well as among different disciplines (e.g., neurologist, psychologist, social worker...) and organizations (general hospital, specialized epilepsy service)
23	Personal support plan	Process and content of support plan
24	Availability and accessibility of support	Sufficient staff, access to support, available support time of experts and staff; reachability of experts
25	Safety and security	Safety of housing and places where support is delivered; includes social safety of the person with epilepsy and support giver(s)
26	Information about epilepsy support services	Information about and familiarity with epilepsy-related support services such as specialized schools, epilepsy teams, epilepsy expert centers
27	Continuity and consistency of support	Turnover of support givers; transfer of support to other experts or support givers (e.g., in case of moving to another area, staff on sick leave, retirement of neurologist)
28	Coordination and management	Coordinating different support resources (e.g., neurologists, psychologists, direct support staff); enhancing support performance; dealing with bureaucracy
<i>Service relationships</i>		
31	Responsiveness	Swiftness of response from support givers to needs and questions
32	Reliability	Trust between person and support givers; keeping promises; providing support as planned; offering support as agreed
33	Communication	Communication between person, support givers, and professionals about the person's health; development of support plan; feedback to person about progress or observed changes in functioning
34	Commitment	Commitment of support givers; motivation; friendliness; spirit; “doing their best”
35	Understanding and empathy	Feeling understood; support giver's sensitivity to the person's needs and emotions

^a Domains 11–18: Schalock et al. [14]; domains 21–28: VGN [15]; domains 31–35: Zeithaml et al. [16].

Proxies completed the survey for the person with epilepsy with whom they had a close relationship as a support giver. They were asked to consider the support that was actually being given to the person with epilepsy.

In the group of 289 respondents, the gender ratio of persons with epilepsy was 51% male and 49% female. Proxy respondent records were significantly more often (60%) related to children, and a significant

proportion (67%) of proxy records was related to persons with intellectual disability. Proxy response was also related more frequently (57%) to persons living with their parents compared with records of respondents with epilepsy who completed the questionnaire independently.

Table 3 shows the relative prevalence of each source in the answers of the respondents. Proxies reported considerably more use of multiple resources than persons with epilepsy.

Table 2

Respondents' characteristics.

	Respondents with epilepsy	Proxy respondents for persons with epilepsy	Total	
	N	N	N	%
Number of respondents	114	175	289	100
Gender of person with epilepsy, m/f	51/63	96/79	147/142	51/49
Age of person with epilepsy				
<18	3	106	109	38
19–30	17	29	46	16
31–60	39	28	67	23
61+	55	12	67	23
Living situation of person with epilepsy				
Living with parents or family member	15	100	115	40
Independently living	66	7	73	25
Residential service	6	52	58	20
Other	27	16	43	15
Impairments of person with epilepsy (patients may be suffering from more than 1 impairment)				
Sensory	10	33	34	12
Physical (motor, cerebral palsy)	18	78	96	33
Intellectual disability	5	117	122	42
Mental health issue – behavioral problem	9	48	57	20
Chronic illness	22	35	57	20
Other	8	16	24	8

Living situation: “Other” – respondents with epilepsy mentioned: ‘with my family/partner/child’... – proxy respondents mentioned: ‘combination of/alternately independent living or in residential facility and with family’ (e.g., weekends or for other periods of time).

Multiple disabilities: “Other” – respondents with epilepsy mentioned: “migraine”, “diabetes”... – respondents from social network mentioned: “PDD-NOS”, “autism”.

Table 3
Prevalence of sources of support used by respondents.

Support source	Respondent group Persons with epilepsy (N = 114)		Respondent group Proxies (N = 175)	
	% of total listings	Number of times mentioned	% of total listings	Number of times mentioned
Specialized epilepsy service	14%	37	22%	252
Residential service (not specialized)	14%	37	18%	206
Professional in home care	9%	23	4%	45
Informal by family/significant other	34%	86	37%	420
Informal by volunteer	4%	11	3%	34
Other	25% ^a	63	16% ^b	190
Total listings	100%	257	100%	1147

^a Other: professionals working in private practice (neurologist, psychologist, social worker); self-help group.

^b Other (proxy): professionals working in private practice (social worker, physiotherapist, teacher); general or academic hospital; hired helper funded by personal budget.

3.2. What do persons with epilepsy report about what they consider to be “good” and what they consider to be “areas of improvement”?

3.2.1. Analysis of qualitative statements

The open response questions resulted in a total of 1176 statements: 704 related to what was experienced as being “good support” and 472 to “areas of improvement”. To organize these results, the content of these statements was first analyzed by coding each answer according to the domains shown in Table 1.

Table 4 shows the relative frequency of statements categorized into 21 domains. Relative presence of statements with respect to “good” or “areas of improvement” domains is shown for the respondents with epilepsy (self), for proxies, and for both groups combined including total response (“good” + “improvement”).

In all groups, statements reflecting positive experiences (“good”) outnumber statements on “improvement”. It is noticeable that the “areas of improvement” category is present more often in the facilitating condition domains in self-report as well as proxy statements. Both groups frequently express satisfaction with the competence of

professional support givers. Proxies mention access and availability of support more often as a problem area. Both groups frequently appreciate support in the domains of commitment and understanding and empathy. The most frequently mentioned domains that, taken together, account for about 50% of all statements are as follows: competence of support giver; access and availability of support; commitment of support givers; understanding and empathy from support givers; and coordination and management of support.

3.3. What do focus groups of persons with epilepsy and their proxies formulate as values of person-centered, long-term support?

On the basis of relative importance of domains as shown in Table 4, vignettes were drafted to summarize what persons with epilepsy perceive as being important in long-term support. These vignettes were evaluated and approved by the discussion panel. The focus groups were then invited to discuss them on the basis of the following questions: ‘are these vignettes recognized in your personal experiences with support?’; ‘do they represent important values?’; and finally,

Table 4
Relative frequencies of domain presence in statements of self and proxy respondents (“good” and “areas of improvement”).

Respondent group	Self	Self	Proxy	Proxy	Self + Proxy	Self + Proxy	Self + Proxy
	“Good”	“Improvement”	“Good”	“Improvement”	“Good”	“Improvement”	Total
Domain	%	%	%	%	%	%	%
1. Quality of life – content related	41	33	29	13	33	16	26
11 Physical well-being	2	2	4	1	4	1	3
12 Emotional well-being	3	2	3	1	3	1	2
13 Interpersonal relations	14	7	4	1	7	2	5
14 Community participation	8	6	5	2	6	3	5
15 Personal development	0	1	1	1	1	1	1
16 Material well-being	8	6	4	2	5	3	4
17 Self determination	5	3	4	2	4	3	3
18 Rights	1	5	4	2	3	3	3
2. Facilitating conditions	30	45	33	67	32	63	45
21 Competence of support givers	17	9	13	8	14	8	12
22 Collaboration between professionals	4	6	4	6	4	6	5
23 Personal supports plan	0	0	3	1	2	1	2
24 Access and availability of supports	4	6	4	28	4	24	12
25 Safety and security	0	3	2	1	2	1	2
26 Familiarity with epilepsy support services	0	14	0	4	0	6	2
27 Continuity and consistency in supports	0	0	4	5	3	4	3
28 Coordination and management	6	7	2	14	3	13	7
3. Service relationships	29	22	38	20	35	20	29
31 Responsiveness	3	9	4	5	4	6	5
32 Reliability	2	3	2	2	2	3	2
33 Communication	2	2	7	7	6	6	6
34 Commitment	8	0	18	3	15	2	10
35 Understanding and empathy	15	7	6	3	9	4	7
Total	100	100	100	100	100	100	100
n statements	195	86	509	386	704	472	1176
n respondents	114	114	175	175	289	289	289
Average n of statements per respondent	1.7	0.8	2.9	2.2	2.4	1.6	4.1

'what are the most important patient values in long-term care and how would you prioritize these?'

Example of a "competence of support givers" vignette:

Persons with epilepsy want to count on the professional's knowledge about their particular type of epilepsy with respect to diagnosis, medication, symptoms, behavioral implications, ability to explain what is going on and competence to undertake appropriate action. Openness is appreciated in talking about "my" epilepsy, not "the" epilepsy. Practitioner's ability to listen to persons with epilepsy and their significant others (partners, parents, children) with respect to epilepsy-related experiences. Experiencing knowledgeable engagement with the professional.

Example of an "emotional well-being" vignette:

Understanding and support in the area of mental health issues, stress management and epilepsy-related behavior problems. Support giver's attention to behavioral problems of persons with multiple disabilities. Privacy and personal space in residential living arrangements. Specific attention to PDD-NOS, autism spectrum disorder, intellectual and developmental disabilities. A holistic approach to the life of the person with epilepsy. Central theme in support should be: reducing uncertainty. Promoting mental fitness in daily life.

Detailed minutes were made of each focus group meeting. Functional analysis and summarizing of the focus groups output resulted in a different structure of topics than the domain categories shown in Tables 1 and 4. There were two reasons for this. In the first place, all focus groups insisted that certain topics were not independent and, therefore, should be seen as strongly interrelated values. Second, the domains were seen as artificial constructs and a grouping, more in alignment with real life experiences of persons with epilepsy, was advised to better represent support values from the person's perspective.

The strongest relation was expressed between professional competence, personal commitment, and an orientation towards the person's well-being that extends to all areas of the person's life and functioning. In the view of the focus groups, these three topics form a cluster and should not be separated.

Another cluster comprised mental fitness, treatment of behavior problems, and emotional well-being of those involved in supporting the person with epilepsy, whether they be a professional or an informal support giver in the person's social network. Coordination of support and availability formed the next clusters of topics.

3.4. Support values through the eyes of the patient: a value framework

After two iterations of comments from the discussion panel, a final format emerged. The outcomes of the focus groups could be organized around four major value clusters, each comprising three interrelated domains that are important to the patient.

3.4.1. Cluster I: relationship between support giver and person with epilepsy

The relationship between support giver and person with epilepsy was most frequently mentioned as an important value. Relationship refers to a dialogue, a two-way communication, in professional consultation settings but also in interactions with nonprofessional support givers in daily life. Three topics were formulated as important.

1. *Knowledgeability and competence* of support givers related to the specific type of epilepsy: the person needs to be confident that the professional (neurologist, nurse, psychologist, direct support giver, informal helper) is informed about his or her specific type of epilepsy and is familiar with its implications, such as symptoms and risks, and is competent to respond adequately. Knowledgeability relates to aspects of the diagnosis, types of seizures, behavioral impact and medication (side-) effects. The need for familiarity with specific characteristics of the particular type of epilepsy extends to

nonprofessional support givers in the direct environment of the person: for example, it is essential that a mother knows how to recognize a seizure in her child (which, as was demonstrated in the focus groups, may not always be obvious).

2. *Empathy and commitment*: persons with epilepsy expect sensitivity to their feelings, needs, and wants when communicating with support givers. Support givers should respect the person's experiences and should be willing and able to adopt his or her perspective. This also implies that support givers are sensitive to how their communication and actions affect the person. Proactive attitude and action are appreciated, for example, enquiring about the well-being of the person outside formal consultation moments or actual support activities. The person expects more than functional commitment from the support giver and values personal commitment to his or her well-being.
3. Support givers should apply a *holistic approach* to the functioning of the person with epilepsy. This implies that in consultation, coaching, treatment, and support situations, various aspects of functioning of the person need to be addressed and not only epilepsy-related issues. A broad perspective implies assessment of support needs in such areas as the person's functioning and role at home, in the family, at school, at work, in leisure settings, in social relationships, as well as the person's physical and mental health and development. In particular, attention should be paid to the effects of epilepsy and medication (1) on health and sleep and (2) on social functioning (relationship with partner, parents, children, colleagues, and friends).

3.4.2. Cluster II: emotional well-being

Epilepsy as a long-term disability is accompanied by feelings of uncertainty and insecurity. Planning for participation in events is often difficult because of the unpredictability of the epilepsy. One never knows when the next seizure will occur or how one will feel afterwards. Vicissitudes of life can make what is easy to achieve on one day hard to achieve on another. This may create irritation and even disbelief in people around the person. In the case of intellectual disability, communication problems or mental health disorders not being understood may contribute to high levels of stress in the person. Feelings of uncertainty are influenced by life phase (adolescence, older age) and life events such as moving or changing school or jobs; they can also be affected by medication. Three topics were formulated as important.

4. Support givers should recognize the person's basic desire for the *pursuit of happiness* and to be a valued person. Support should enhance mental fitness, stress control, and feelings of safety and security. Sources of stress and frustration should be recognized at home as well as in the classroom or at work. During the day, there should be sufficient opportunity for relaxation. In residential services, group living should not be underestimated as a source of stress. Support givers should take a positive attitude, focusing on life-satisfaction instead of the person's disabilities and handicap.
5. Support should include attention to *associated challenges* such as autism spectrum disorder, learning disabilities, intellectual disabilities, mood disorders, behavior problems, memory problems, and mental health issues, such as bipolar disorder and schizophrenia. If such problems are present, professional advice should be integrated into the support activities.
6. *Emotional well-being of support givers* themselves is a value to be observed in order to optimize the support process. Balance between capabilities and workload is an important condition for both professional and informal carers in long-term support. Burn-out risks and early signs should be recognized and responded to in order to maintain support capacity and quality.

3.4.3. Cluster III: coordination of support

Long-term support of persons with epilepsy implies a significant number of arrangements, planning activities, and bureaucracy. Making appointments, applying for services, referrals, keeping records,

communication with health insurance, and municipal services demand a lot of time and skills from the person with epilepsy as well as from support givers. Fluctuations in health condition – typical in epilepsy – as well as in contextual circumstances of the person add to the complexity and to the importance of good coordination measures. Three topics were seen as being especially important.

7. *Coordination of information* between professionals involved in the person's support, especially in the collaboration of experts within a team, in the course of a referral and when different organizations are involved. Professionals should draw on the same information sources and enhance consistency in their communication. Coordination of relevant information should also occur between professional and informal support givers and among informal support givers.
8. *Continuity and consistency in actions* between support givers enhance trust and reliability, factors which require attention particularly in situations of transition, for example: transition from school to work; moving to another place; retirement of the person's familiar neurologist; and changing from child services to adult services. Staff discontinuity in professional services is another risk for discontinuity and loss of reliability of support.
9. *Coordination of practical issues* such as applying for services, accessing funding or subsidies, finding reimbursement for aids and devices, filling in forms, or organizing transportation: rules may differ from one organization or local authority to another. Coordination of systems and between organizations or authorities reduces the burden of bureaucracy.

3.4.4. Cluster IV: access to support services

Availability, accessibility, and reachability of support services and networks are important structural conditions underlying efficacy of the previous three value clusters. The following topics were formulated by the focus groups.

10. *Aids*, such as specialized e-health apps as well as generic apps to enhance planning, communication, and organization within the support network, are seen as important technical facilitators. The same is true for alarm devices, safety devices, and early warning systems. Technical devices and Information and Communications Technology (ICT) applications can enhance health, safety, and trust, and their relevance and potential contribution should be assessed in every individual support plan.
11. *Social networks* of parents, partners, and children and networks of informal as well as professional support givers are important conditions for enhancing effective support. Contact between persons with epilepsy and between informal support givers with people in similar situations is seen as an important characteristic of sound support. Membership of and involvement in collective advocacy associations are mentioned as important to persons with epilepsy and their significant others. Such associations are important in promoting knowledge about epilepsy and the inclusion of persons with epilepsy in society at large.
12. *Reachability* of specialized services is important to persons with epilepsy. Travel distances and travel time to a special service such as a school or epilepsy center are seen as hindrances to good support. Swift access to specialists (such as neurologists) – for example, contacting a specialized service from a general hospital – is not evident but may significantly enhance perceived treatment and support quality, especially in emergency situations or when there is doubt about diagnosis or treatment.

3.5. Reflective questions for patients, informal support givers, and professional support givers resulting from the value framework

It was intended that the value framework should be useful in practice, for instance, for validating and operationalizing actions and

instruments to promote a person-centered focus in long-term support for persons with epilepsy. An initial draft of items was formulated by the first author and presented to the discussion panel for validation against the framework and then to reach agreement about the formulation. After two iterations, consensus was reached on three sets of questions that translate the value domains into relevant items in practice. The addendum presents the translation of the value framework into reflective questions for (1) the person with epilepsy, (2) the informal support giver, and (3) the professional support giver in order to enhance awareness of relevant person-centered issues in long-term support. These sets of questions represent an exemplary translation of the value framework that can be used for example in educational settings or team evaluation in service settings to raise awareness about person-centered support.

4. Conclusion and discussion

Knowledge about what elements of long-term support are positively valued by persons with epilepsy is relevant to enhance person-centeredness in interventions and assistance, both by professional and informal carers. The present study led to formulating twelve value domains of supports grouped into four clusters (Section 3.4). The study started with a survey-collected statements about experiences of “good support” as well as of “areas of improvement” in the respondents' own wordings. Statements were next organized and summarized in vignettes of frequently recurring subjects and presented to focus groups of persons with epilepsy and long-term support needs and to an expert group of stakeholders. After four iterations, consensus was reached on twelve value domains. Next, the twelve value domains were operationalized into reflective questions for persons with epilepsy, their informal carers, and for professionals (Section 3.5). These questions (see Appendix A) can be used as a checklist for self-examination with respect to person-centeredness of support as offered to people with epilepsy. They may also be useful in education, training, and team evaluation settings.

Some value domains in the present study are congruent with findings from Kalsbeek and Plateel's Dutch study into quality criteria from the patients' perspective for professional epilepsy care in early diagnosis and initial treatment. Of their seven criteria, five items were also found in the present study on long-term support. These refer to the importance of (1) epilepsy-specific knowledge and competence, (2) an integral approach to the patient's health, (3) a healthy network of significant others and involved support givers around the person with epilepsy (beware of support burden risks), (4) collaboration between professionals from various disciplines involved (within a basic epilepsy team), and (5) recognizing the added value from patients' organizations. The two other topics in the Kalsbeek and Plateel study concern the individual epilepsy support plan and specific attention to continuity of medication strategy and management. The personal support plan was not specifically reported as a support value in the present study. Continuity was reported as a value, though it was extended beyond medication and applied to continuity of support at large including consistency between practitioners.

The present study of long-term support emphasizes additionally (1) the importance of personal commitment and empathy in relationships between support givers (whether or not professionals) and the person with epilepsy; (2) the importance of a wide-angle view of the impact of epilepsy on human functioning, which calls for the use of models such as the WHO International Classification of Functioning (ICF), the supports model [9], or models of quality of life (for example, [14,17]) as diagnostic aids; (3) the importance of the person's mental fitness and emotional well-being, including specific attention to the unpredictable nature of epilepsy, behavioral problems, and mental health issues or comorbidity; (4) the importance of the mental health and fitness of the support givers; (5) the importance of continuity of support throughout transitions and changes of specialists, support staff, or other persons

involved; (6) the importance of accessibility of services (generic such as municipality services, basic health services, community activities, as well as specialized services from expert centers); and (7) the importance and potentials of technical and electronic aids for communication and monitoring.

Person-centeredness is now widely accepted as a core feature of quality of healthcare [18–20], including long-term care and support for persons with chronic illness or impairments [21–24]. Gerteis et al. [25] defined dimensions of person-centered care that were later empirically confirmed [26] and comprised of the following: (1) respect for patients' values, preferences, and expressed needs; (2) coordination of care; (3) information, education, and communication; (4) physical comfort; (5) emotional support; (6) involvement of family and friends; and (7) transition and continuity.

It is obvious that there is overlap between person-centered values in long-term epilepsy support, as found in the present study, and characteristics of person-centered healthcare at large, as defined in other studies. More specific findings in the present study relate to the following: (1) the fact that patient values in long-term support care do not fundamentally differ between support received from professionals and from informal support givers; (2) there are no obvious differences regarding long-term support values across subgroups of persons with refractory epilepsy (comorbidity, living situation); (3) the importance of knowledgeability of the support giver about the specific type of epilepsy; (4) the sensitivity and responsiveness of the support giver to the unpredictability of refractory epilepsy; (5) the importance of personal commitment and empathy in relationships between persons with epilepsy and support givers; (6) the importance of accessibility of services (generic services such as municipality services, basic health services, community activities, as well as specialized services from expert centers); and (7) the importance and potentials of technical and electronic aids for communication and monitoring epilepsy-related risks.

Our project was set up as a practical study and used a convenience sample of 289 persons with epilepsy, 179 of whom were represented by proxies. In quality-of-life research, it is known that proxy responses should be interpreted with caution, and data should be treated

separately [27–29]. However, careful instruction of proxies significantly enhances reliability of proxy responses [30]. In the present study, special attention was given to the instruction of proxies both in the survey and in the focus groups, and distinction was made between data sources (persons with epilepsy and proxies).

The present study used qualitative data of respondents' statement about their experiences with support related to having epilepsy. Classification of these data was done by reading and coding each statement according to an existing quality-of-care frame of Dutch service providers in disability care and summarizing the most frequently stated subjects in each category into vignettes for discussion by focus groups. This procedure is congruent with the involvement of stakeholders in a dialogue to reaching consensus on the intended value domains. However, the use of new techniques of big data analysis such as Word Adjacency Graph Modeling (WAGM) – using, for example, data from specialized patient help desks or patient advocacy forums – may offer an alternative methodology in identifying patient values [31]. Although WAGM analysis requires much greater numbers of data than were available in the present study, its use in future similar research projects should be considered.

Since the present study is clearly based on experiences and appraisals of persons who are familiar with the Dutch epilepsy care system, results should be regarded as representing this particular situation. No validity is claimed beyond this population. However, a comparison of our findings with outcomes of international studies, mentioned above, shows congruencies that could be indicative of more general values in long-term care in other jurisdictions and other long-term healthcare areas. Systematic comparison and reviews of such studies in the field of (long-term) epilepsy may reveal more universality in patient values which should be taken into account in person-centered long-term care.

Acknowledgment

This study was funded by a grant from the Ds. Visscher Foundation (Groesbeek, the Netherlands) and the Dutch Epilepsy Fund.

Appendix A. Addendum

Reflective questions to evaluate long-term support situations from the perspective of (1) a person with epilepsy, (2) an informal support giver, and (3) a professional support giver.

Value cluster	Domain	Self-examination items for		
		(1) The person with epilepsy	(2) The informal support giver	(3) The professional support giver
Relationship between support giver and person with epilepsy	1 <i>Knowledgeability and competence</i>	Am I confident that professionals and informal support givers whom I deal with are informed about my specific type of epilepsy? What would I like to improve?	Since there are many types of epilepsy, it is important to know the characteristics and risks of the specific type of epilepsy of the person whom I am supporting. Do I know enough about the case of the person whom I support? What can I do to improve?	Do I know about the specific type of epilepsy of the individuals whom I support? Am I able to give specific information about their epilepsy type and seizure types? Am I able to give tailored advice or to refer to qualified experts about therapeutic options, including medication, surgery, etc.? Do I inform the patient and his/her support givers about associated risks and side effects of medication? What could I improve?
	2 <i>Empathy and commitment</i>	Do I feel that professionals and informal support givers are committed to supporting me? Do I feel understood? Do they care? What would I like to improve?	Do I feel committed to the person(s) with epilepsy whom I support? Do I understand the needs and wants of the person(s) I am interacting with? What can I do to improve?	Am I sensitive to the support needs, feelings, and wishes of persons with epilepsy whom I am supporting? Do I monitor the effects of my communications and interventions? Do I show interest in their well-being, also between consultations?
	3 <i>Holistic view</i>	Do I feel that professionals and informal support givers are interested in my well-being in a broader sense and not just taking a technical interest in epilepsy symptoms or seizure count? Do they take into consideration the impact of my	Do I look at the functioning of the person with epilepsy as a person and not just as an epilepsy patient? Am I sensitive to what type of support may contribute to his or her physical and emotional well-being, social life (family, school, work, relationships,	In my interventions as a professional, do I discuss with the patient his or her support needs in the broad area of human functioning? Does my perspective transcend a narrow focus on epilepsy as impairment? Does my professional

(continued on next page)

Appendix A (continued)

Value cluster	Domain	Self-examination items for		
		(1) The person with epilepsy	(2) The informal support giver	(3) The professional support giver
		epilepsy and medication on my physical and emotional well-being, my social life (family, school, work, relationships, mobility), development, material well-being, rights? What would I like to improve?	mobility), development, material well-being, rights? Am I in a position to contact or refer to support sources that may be beneficial? What can I do to improve?	assessment perspective include attention to the patient's physical and emotional well-being, social life (family, school, work, relationships, mobility), development, material well-being, rights? Am I in a position to contact, engage, or refer to specialists from relevant disciplines in order to enhance the support for this patient?
Emotional well-being	4 Pursuit of happiness	Do I feel that my present support givers pay sufficient attention to my emotional well-being? Are my self-determination and choices respected? Do I feel supported in coping with behavioral and mental effects of epilepsy and my medication? How do I cope with stress? Do I have moments that I can sit back and relax? What would I like to improve?	Do I pay attention to the emotional well-being of the person(s) whom I support? Do I seek to recognize and reduce stress in the person and to encourage self-determination and choice? Am I aware of mental health risks of the person? What can I do to improve?	As a professional, do I pay attention to the emotional well-being of the person(s) whom I am supporting? Do I seek to recognize and reduce stress in the person and to enhance self-determination and choice? Am I aware of mental health effects and risks of the specific medication of the patient? If epilepsy and/or related medication do not explain certain mental health issues, do I refer to specialized colleagues for advice or further diagnosis and treatment? How can I improve my professional working methods?
	5 Associated challenges	Epilepsy may be accompanied by mental health problems or disorders such as autism spectrum disorder, intellectual or developmental disabilities, PPD-NOS, ADHD, or other challenges. If applicable, do I feel that there is special attention to such problems in the support I experience from professionals as well as informal support givers? What would I like to improve?	Epilepsy may be accompanied by mental health problems or disorders such as autism spectrum disorder, intellectual or developmental disabilities, PPD-NOS, ADHD, or other challenges. Do I feel I know enough about such problems and do I know what they may mean for my support activities? Do I know how to turn to professional help if I feel this is desirable or necessary? What can I do to improve?	Epilepsy may be accompanied by mental health problems or disorders such as autism spectrum disorder, intellectual or developmental disabilities, PPD-NOS, ADHD, behavior problems, or other challenges. Does this patient receive special attention for these problems? Do I know what implications there may be for my own approach to the patient's treatment and support? Do I know how to contact relevant experts for my own information or for referral?
	6 Well-being of support givers	It is important that people whom I live with and rely on for support (whether professionals or not) are sufficiently fit for their task. Do I experience that they are? Do I trust them in their capability and fitness to offer the assistance that is required? What would I like to change?	I feel sufficiently fit to support the person (s) who needs my help. I try to keep myself fit for my duties and to avoid burnout so that I can continue my support activities in a healthy way. I can refer to assistance should I feel less able to deliver the support expected from me. What can I do to improve?	What should be improved in this respect? Helping other people can be stressful for professionals as well as for informal support givers in settings such as the family, school, work, and residential care. Do I, as a professional, pay attention to the burden on those who support patients with epilepsy? Do I pay attention and do I take responsibility to avoid burnout in professional and informal carers? What could I improve?
Coordination of support	7 Coordination of relevant patient information	How do professionals who are involved in my treatment and support work together? Are they informed about my support needs? Do they exchange information? Is their advice and are their interventions consistent? What would I like to improve?	Do I communicate with other informal support givers? Do I align my support with activities by others (informal support givers as well as professionals)? What can I do to improve?	Do I check if my contribution is aligned with support and interventions from other professionals? Are interventions coordinated? Do professionals share the same information and data sources to support the patient? What could I improve?
	8 Continuity and consistency	Is there continuity in my supports? Do support givers (specialists, staff) change a lot? Do I have concerns regarding (upcoming) transitions in my life? What would I like to improve?	Do I provide for continuity in the supports of the person? Do I try to avoid inconsistency in supports? What can I do to improve?	Do I as a professional contribute to smooth transitions from one care situation to another? Do I promote continuity and consistency in interventions and supports? Are measures taken to minimize staff changes and promote team stability? If changes cannot be avoided, do I do my best to ensure continuity of support? What could I improve?
	9 Coordination of practical issues	How do I experience support in planning and handling such things as making appointments, using transportation, filling in forms, applying for devices, making arrangements? What would I like to improve?	How well am I managing to assist the person with planning and handling his or her diary, making appointments, organizing transportation, filling in forms, applying for services and making arrangements? What would I like to improve?	Do I pay sufficient attention to facilitating practical and bureaucratic implications of my interventions (including planning, transportation, applying for services)? Do I refer matters efficiently in order to avoid overburdening the patient and his or her supportive network? What could I improve?
Access to support	10 Aids	Might technical aids and apps be useful for me, for example, to contact a practitioner or support person (e-health)? As a safeguard? What would I like to improve?	Technical aids and apps may be useful in facilitating my tasks as a support giver. What would I like to enquire about?	Am I aware of technical apps and aids that might facilitate communication with the patient and his or her supportive network (for example, E-Health solutions)? Am I

Appendix A (continued)

Value cluster	Domain	Self-examination items for		
		(1) The person with epilepsy	(2) The informal support giver	(3) The professional support giver
	11 <i>Social networks</i>	Am I aware of patient associations and self-advocacy groups? Am I a member? Do I receive information about meeting others in the same situation and learn from their experiences? Am I able to retrieve information from websites of patient associations and epilepsy centers? What would I like to improve?	Do I meet with other informal support givers to exchange information, to learn from them, and to help them with my own experience? Am I a member of an epilepsy association or a self-help group? Do I receive information from epilepsy associations and expert centers about relevant developments in treatment, policy, organization, and procedures? What can I do to improve?	aware of the importance of risk management and aids that facilitate seizure detection or early warning? Is technology considered as a resource in the patient's individual support plan? What could I improve?
	12 <i>Reachability</i>	Where I live, do I have reasonable physical access to epilepsy services (travel time)? Do I have reasonable access to an epilepsy team (neurologist, specialized nurse practitioner, other experts)? Do I experience understanding and support from school, work, local health services, and public services (municipality and community)? Do I have access to and do I receive adequate support to enhance my participation in the community? What would I like to improve?	Do I experience understanding and support from school, work, public services (municipality and community), and health services with respect to my role as a support giver of a person with epilepsy? With respect to my role in supporting the person with epilepsy: do I have easy access to practitioners in case I need information or help? What would I like to improve?	Do I have information about patient associations and self-help groups in epilepsy? Can I give advice to patients on how to find or to join such associations and participate in activities of persons in the same situation? Do I know the social map of epilepsy resources in my region? What could I improve? As a professional, am I sensitive to the role that environments such as epilepsy expertise centers, school (teachers), work (employers, colleagues), and leisure (sports clubs) play in the life of my patient(s)? Am I aware of the impact that public services such as police, health services, ambulance personnel, and attitudes of the public have on the functioning of persons with epilepsy and their informal support givers? Do I (personally or as member of a team or organization) contribute to the functioning of persons with epilepsy in the community? What could I improve?

References

- [1] Sillanpää M, Schmidt D. Long-term outcome of medically treated epilepsy. *Seizure* 2017;44:211–6.
- [2] Kalsbeek C, Plateel V. Epilepsie. Kwaliteitscriteria vanuit patiëntenperspectief. Ede: Epilepsie Vereniging Nederland; 2012.
- [3] Laxer K, Trinkka E, Hirsch L, Cendes F, Langfitt J, Delanty N, et al. The consequences of refractory epilepsy and its treatment. *Epilepsy Behav* 2014;37(2014):59–70.
- [4] Elwyn G, Todd S, Hibbs R, Thapar A, Edwards P, Webb A, et al. A 'real puzzle': the views of patients with epilepsy about the organisation of care. *BMC Fam Pract* 2003;4(4).
- [5] Miller WR, Bakas T, Buelow JM. Problems, needs, and useful strategies in older adults self-managing epilepsy: implications for patient education and future intervention programs. *Epilepsy Behav* 2014;31:25–30.
- [6] Wagner JL, Brooks B, Smith G, St. Marie K, Kellermann TS, Wilson D, et al. Determining patient needs: a partnership with South Carolina Advocates for Epilepsy (SAFE). *Epilepsy Behav* 2015;51:294–9.
- [7] Mahendran M, Speechley KN, Widjaja E. Systematic review of unmet healthcare needs in patients with epilepsy. *Epilepsy Behav* 2017;75:102–9.
- [8] Schalock R, Luckasson R. What's at stake in the lives of people with intellectual disability? Part I: the power of naming, defining, diagnosing, classifying, and planning supports. *Intellect Dev Disabil* 2013;51(2):86–93.
- [9] Thompson JR, Bradley V, Buntinx WHE, Schalock RL, Shogren KA, Snell M, et al. Conceptualizing supports and the support needs of people with intellectual disabilities. *Intellect Dev Disabil* 2009;47(2):135–46.
- [10] Porter M. What is value in health care? *N Engl J Med* 2010;363:2477–81.
- [11] Ritchie J, Lewis J, McNaughton Nicholls C, Ormston R, editors. Qualitative research practice. A guide for social science students and researchers. Los Angeles: Sage; 2014.
- [12] Widdershoven GAM. Dialogue in evaluation: a hermeneutic perspective. *Evaluation* 2001;7(2):253–63.
- [13] Abma T, Widdershoven G. Responsieve methodologie. Interactief onderzoek in de praktijk. Amsterdam: Boom [Responsive methodology. Interactive research in practice]; 2006.
- [14] Schalock RL, Bonham GS, Verdugo MA. The conceptualization and measurement of quality of life: implications for program planning and evaluation in the field of intellectual disabilities. *Eval Program Plann* 2008;31(2):181–90.
- [15] VGN Vereniging Gehandicaptenzorg Nederland. Visiedocument Kwaliteitskader Gehandicaptenzorg [quality of disability care framework] 2.0. Utrecht: VGN; 2013.
- [16] Zeithaml VA, Bitner MJ, Gremler DD. Services marketing: integrating customer focus across the firm. New York: McGraw-Hill; 2006.
- [17] Buntinx WHE, Schalock R. Models of disability, quality of life, and individualized supports: implications for professional practice in intellectual disability. *J Policy Pract Intellect Disabil* 2010;7(4):283–94.
- [18] Davis K, Schoenbaum SC, Audet AM. A 2020 vision of patient-centered primary care. *J Gen Intern Med* 2005;20(10):953–7. <https://doi.org/10.1111/j.1525-1497.2005.0178.x>.
- [19] Epstein RM, Street M. The values and value of patient-centered care. *Ann Fam Med* 2005;9(2):100–3. <https://doi.org/10.1370/afm.1239>.
- [20] Institute of Medicine. Crossing the quality chasm: a new health system for the 21st century. Washington, DC: National Academy Press; 2001.
- [21] Cramm JM, Leensvaart L, Berghout M, Exel, van, J. Exploring views on what is important for patient-centred care in end-stage renal disease using Q methodology. *BMC Nephrol* 2015;28(16):74. <https://doi.org/10.1186/s12882-015-0071-z>.
- [22] Frampton S, Guastello S, Kinsey J, Boudreau-Scott D, Lepore M, Henfey A, et al. Long-term care improvement guide. Derby (CT)/Camden (ME): Planetree Inc. & Picker Institute; 2010.
- [23] Lin JD, Hu J, Yen CF, Hsu SW, Lin LP, Loh CH, et al. Quality of life in caregivers of children and adolescents with intellectual disabilities: use of WHOQOL-BREF survey. *Res Dev Disabil* 2009;30:1448–58. <https://doi.org/10.1016/j.ridd.2009.07.005>.
- [24] Miller NA. Patient centered long-term care. *Health Care Financ Rev* 2010;19(2):1–10.
- [25] Gerteis M, Edgman-Levitan S, Daley J, Delbanco TL. Through the patients' eyes: understanding and promoting patient-centered care. San Francisco, CA: Jossey-Bass; 1993.
- [26] Rathert C, Williams E, McCaughey D, Ishqaidif G. Patient perceptions of patient-centred care: empirical test of a theoretical model. *Health Expect* 2015;18(2):199–209.
- [27] Schmidt S, Power M, Green A b, Lucas-Carrasco R. Self and proxy rating of quality of life in adults with intellectual disabilities: results from the DISQOL study assessment strategies. *Res Dev Disabil* 2010;31:1015–26.
- [28] Stancliffe R. Proxy respondents and quality of life. *Eval Program Plann* 2000;23(1):89–93.
- [29] Verdugo MA, Schalock RL, Keith KD, Stancliffe R. Quality of life and its measurement: important principles and guidelines. *J Intellect Disabil Res* 2005;49(10):707–17.
- [30] Balboni G, Coscarelli A, Giunti G, Schalock RL. The assessment of the quality of life of adults with intellectual disability: the use of self-report and report of others. *Res Dev Disabil* 2013;34:4248–54.
- [31] Miller WR, Groves D, Knopf A, Otte JL, Silverman RD. Word adjacency graph modeling: separating signal from noise in bog data. *West J Nurs Res* 2017;39(1):166–85.