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Support needs of caregivers of patients with amyotrophic lateral sclerosis: A qualitative study

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Abstract

Objective. The aim of this study was to explore the support needs of Dutch informal caregivers of patients with amyotrophic lateral sclerosis (ALS).

Method. Individual semi-structured interviews were conducted with 21 caregivers of ALS patients. Audio-taped interviews were transcribed and data were analyzed thematically.

Result. A total of four global support needs emerged: “more personal time”, “assistance in applying for resources”, “counseling”, and “peer contact”. Despite their needs, caregivers are reluctant to apply for and accept support. They saw their own needs as secondary to the needs of the patients.

Significance of results. ALS seems to lead to an intensive caregiving situation with multiple needs emerging in a short period. This study offers targets for the development of supportive interventions. A proactive approach seems essential, acknowledging the importance of the role of the caregivers in the care process at an early stage, informing them about the risk of burden, monitoring their wellbeing, and repeatedly offering support opportunities. Using e-health may help tailor interventions to the caregivers’ support needs.

Introduction

Amyotrophic lateral sclerosis (ALS) is a fatal neurodegenerative disease that affects motor neurons, leading to the loss of all voluntary muscle function (Mitchell et al., 2007). Although ALS is predominantly known for its physical deterioration, patients may also develop cognitive and behavioral symptoms including apathy, disinhibition, and impairments in executive functioning (Goldstein et al., 2013).

ALS is often described as a family illness because it takes its toll not only on the patients, but also on their immediate social environment (Cipolletta et al., 2015). The majority of patients live at home and most of their care is provided by relatives, primarily partners. These informal caregivers spend increasing proportions of their time on caregiving activities, ranging from physical care to housekeeping tasks (Chio et al., 2006). As a result, caregivers often experience physical symptoms, such as exhaustion, fatigue, and sleeping problems (Ray et al., 2007; Van Teijlingen et al., 2001). ALS not only puts a strain on caregivers in terms of time and energy, this burden also includes a significant psychological component. The close intertwining of their life with that of the patient means that caregivers often feel that their own lives have been turned upside down (Anderson et al., 2016).

Previous research showed that caregivers who faced high care demands were more likely to experience poorer physical and mental health, particularly when they felt they lacked control over their caregiving tasks (Molloy et al., 2008; Orbell et al., 1993). Feelings of depression, psychological distress, and quality of life of ALS caregivers worsen as the disease progresses and care demands increase (Gauthier et al., 2007; Goldstein et al., 2006). Although caregivers face high care demands, not all of them become distressed (Lillo et al., 2012). This would seem to indicate that the feelings of distress perceived by the caregivers are not only influenced by the demands made upon them, but also by other factors, such as control.

Despite the awareness of the decreased physical and mental health of caregivers and the importance of family support in comprehensive ALS patient care, evidence-based supportive interventions for caregivers are lacking (Aoun et al., 2013). A first step in the development of such interventions for ALS caregivers is to identify their support needs when caring for the

patient. Qualitative research into this topic has previously been conducted in other countries. A study in the United Kingdom showed that caregivers reported a significant need for training in the manual handling and physical care of the patient (O'Brien et al., 2012). A Swedish study reported the need for caregiver support groups to exchange experiences with caregiving, as well as a need for information and knowledge about the disease and care, and a need for bereavement support (Larsson et al., 2015). In an Australian study, caregivers indicated they needed support to cope with experiences of loss and to adapt to changes in the care situation during the course of the disease (Anderson et al., 2016). Care needs seem to differ across countries, and these differences may be caused by cultural differences and differences in the organization of healthcare and support services (Bede et al., 2011).

In the Netherlands, care is organized along three disease phases: the diagnostic, rehabilitation, and terminal phases (Van den Berg et al., 2004). The diagnostic phase is coordinated by a neurologist, the (palliative) rehabilitation phase by a rehabilitation physician, and the terminal phase by a general practitioner. In the rehabilitation phase, care is provided by a multidisciplinary ALS care team and the social worker or psychologist of the team can also offer support to caregivers. Because of cultural and healthcare differences, support needs of caregivers in The Netherlands may differ from those of caregivers in other countries. Therefore, the goal of this study was to explore the specific support needs of informal caregivers of patients with ALS in the Netherlands.

Methods

Study design

This study used a qualitative research design based on thematic analysis (Braun et al., 2006). The study was submitted to the Medical Ethics Committee of the University Medical Center Utrecht in the Netherlands (15-272C), which deemed it exempt from review. Participation was voluntary, and all participants signed an informed consent form before the interview. The reporting in this paper is in accordance with the Standards for Reporting Qualitative Research checklist (O'Brien et al., 2014).

Participants

Informal caregivers of ALS patients were recruited via six specialized ALS care teams in hospitals or rehabilitation centers located in different regions of the Netherlands, using random sampling: any caregiver who was seen by the ALS care team could be included in the study. We asked rehabilitation physicians, psychologists, and social workers to inform caregivers about our study by means of an information leaflet. Those interested were contacted and interviewed once. Twenty-one caregivers of patients with ALS were interviewed between February 2015 and August 2015. Table 1 presents the demographic characteristics of those interviewed; most of whom were women ($n = 15$) and co-resident partners of patients ($n = 15$). The majority ($n = 14$) experienced feelings of burden. Ten caregivers were providing care for patients with moderate disabilities, seven for patients with severe disabilities and three for patients who were very severely disabled. Twelve caregivers were interviewed within the first year after the patient's diagnosis.

Table 1. Characteristics of caregivers

Characteristic	$n = 21$
Group	
Current family caregiver	20
Bereaved family caregiver	1
Gender	
Female	15
Male	6
Relationship to patient	
Partner	15
Child	5
Sibling	1
Age	
70–79	3
60–69	4
50–59	6
40–49	2
30–39	4
20–29	2
Country of birth	
The Netherlands	20
Other	1
CSI score*	
0–6 (no burden)	6
7–13 (burden)	14
Time between diagnosis and interview	
<1 year	12
1–2 years	4
3–4 years	4
>4 years	1
ALS-FRS-R score patient*	$M = 22.5$ ($SD = 9.2$)
37–48 (mild physical disabilities)	–
25–36 (moderate physical disabilities)	10
13–24 (severe physical disabilities)	7
0–12 (very severe physical disabilities)	3

ALSFRS-R, Amyotrophic Lateral Sclerosis Functional Rating Scale-Revised; CSI, Caregiver Strain Index.

*These questionnaires were not completed by the bereaved caregiver.

Data collection

Interviews were conducted by the first author in a semi-structured format. The interviewer was not known to the participants before the start of the study. Based on a literature review, we formulated an interview guide including topics related to caregivers' need for support (Appendix 1).

The face-to-face interviews were held at a time and location of the participants' choosing (either in a rehabilitation center or in their own home) and were conducted in a private setting. The patient was not present. The duration of the interviews ranged from 32 to 88 minutes. To gain insight into the severity of the patient's disabilities and the care demands placed on the

caregiver, we asked participants to complete the Amyotrophic Lateral Sclerosis Functional Rating Scale-Revised (ALSFRS-R) (Cedarbaum et al., 1999). This is a validated questionnaire-based scale that assesses the physical functioning of ALS patients, including gross motor tasks, fine motor tasks, bulbar functions, and respiratory function. Higher scores denote better physical functioning. To facilitate clinical interpretation of the findings, the ALSFRS-R scores were categorized into four stages of severity: mild (37–48), moderate (25–36), severe (13–24), and very severe (0–12) (Mioshi et al., 2012). We also asked caregivers to complete the Caregiver Strain Index (Robinson, 1983), a questionnaire assessing strain as a result of caregiving. It consists of 13 items; total scores range from 0 to 13, with 7 or more indicating caregiver strain. It is an instrument with good construct validity and internal consistency, which can rapidly identify individuals with potential caregiving concerns (Robinson, 1983). After the interviews, respondent validation of the acquired data was performed by member checking (Creswell et al., 2000).

Data analysis

Interviews were audio-recorded, transcribed, anonymized, and subsequently analyzed by two researchers (JdW, JeM). The process from data collection to analysis was iterative because these processes were carried out simultaneously, so important themes that emerged from the analysis could be incorporated into subsequent interviews until saturation was reached (Guest et al., 2006). Data saturation was achieved when no new themes were added during the last three interviews.

The first step of data analysis involved repeated reading of the transcripts to become familiar with the narratives and to annotate what was significant. Next, the texts were broken down into fragments based on content, and these fragments were labelled with codes using NVIVO 10 (NVivo Qualitative Data Analysis Software, 2012). This process was performed by the two researchers independently. Results of their coding were compared and discrepancies discussed to enhance the credibility of the results and to minimize interpretation bias. Once the coding of all interviews had been completed, codes were sorted in terms of similarities and overarching themes and subthemes were identified based on the codes. Finally, a summary was written on each theme through a collaborative effort between the authors. Quotes were linked to the themes that expressed the essence of the content.

Results

Support needs

Four major themes were identified with regard to the support needs of ALS caregivers: “more personal time”, “assistance in applying for resources”, “counseling”, and “peer contact”. These, together with subthemes and quotations, are presented in Table 2. The results are discussed by theme in the following section.

Having more personal time

Many caregivers sacrificed their working hours, leisure activities, and social life to provide care. They adapted their lifestyle to the lifestyle of the patient, which resulted in a more restricted and inactive life (quote 1 in Table 2). Providing care was often considered as burdensome, and having more personal time

would offer some relief. Caregivers indicated that being able to leave the house and spend time on their own activities was important for maintaining their own wellbeing, and allowed them to divert their attention from ALS.

A significant strain, as well as an important barrier to having more personal time, was that many caregivers were constantly on standby for their family member with ALS (quote 2). This could partly be attributed to the continuous care tasks, even with paid home-care. But, an important factor here was the caregivers' concern that, in their absence, something could happen to the patient (e.g., falling, choking). Therefore, leaving the house was no longer a spontaneous activity and needed to be carefully coordinated, since alternative care had to be organized (quote 3). Arranging this could be difficult because people in the caregivers' social network were often not equipped to take over care tasks.

A barrier to requesting (more) professional care was the lack of privacy inside the home because of the presence of healthcare professionals (quote 4). One participant specifically chose to deliver the care by himself to maintain his privacy. Another option to create more personal time was respite care, in which professionals take over the responsibilities temporarily in order to relieve the caregiver. None of the caregivers had actually used respite care, although many had explored this possibility. Most were told that their care situation was too complex for respite care and that the required care needs could not be met (quote 5). In one situation, respite care could only be provided at a nursing facility, which was rejected by the patient. Caregivers indicated that patients (strongly) preferred receiving care from their primary caregiver, making it complicated to get more personal time (quote 6). To deal with the lack of personal time, some caregivers tried to focus on the temporary nature of care, as ALS implies a short life expectancy. This mind-set helped them persevere, but at the risk of continuously asking too much of themselves (quote 7).

Assistance in applying for resources

Caregivers expressed the need for professional help in managing the logistics of care provision. Most participants felt that applying for healthcare insurance reimbursement, home-care, and aids, such as wheelchairs and communication technology, was a lengthy and convoluted process, leaving many frustrated (quote 8). They had to go through different mandatory procedures before the required care could be provided (quote 9). Consequently, some devices were delivered too late, as the condition had already progressed to a stage where the device was no longer useful.

For some caregivers, it was not clear what kinds of devices were available and what type of professional home-care they could receive. Participants expressed a need for information on available support options. They would like to be supported in finding the right contacts and routes to get funding or aids (quote 10).

Counseling

Some caregivers indicated that speaking to a professional in the absence of the patient would provide relief (quote 11) and allow them to speak freely about their own concerns and needs without worrying how their statements might affect the person with ALS (quote 12). Caregivers who discussed their issues with their social network often felt guilty when they talked negatively about the patient or the required care. They stated that counseling might

Table 2. Overview of categories and subthemes of caregiver needs

Categories of ALS caregiver needs	Subthemes	Quotes (respondent number, gender, age)
More personal time	• Restricted personal time due to caring	1. "Spare time, I don't really have that. That's just how it is when you provide care to someone who can't do anything. I would like to do a lot of things but I'm limited." (R17, male, 59) 2. "Continuously being available. I find that very difficult (...) I feel like I'm only taking care of the basic things in life. There's no room for relaxation." (R3, female, 47) 3. "People say 'You have to do some fun things, you have to think of yourself.' I understand that, but that's actually quite complicated." (R9, female, 35)
	• Lack of privacy	4. "There are constantly people in our home; I don't have a private life anymore." (R4, male, 73)
	• Respite care	5. "Respite care is too complicated in our family. I have been in contact about this topic several times. But it's only possible if ... and that doesn't cover our situation." (R3, female, 47) 6. "Yes, I would like it [respite care] very much. But I guess my father thinks otherwise." (R6, female, 38)
	• Focus on temporariness	7. "I believe that she will live one more year. I can get through that year." (R2, male, 71)
	Assistance in applying for resources	• Aids 8. "It would be so helpful if the team [rehabilitation team] would apply for the devices for me. It would really help my frustration. I put a lot of time and effort in it [applying for devices] but I'm just not getting anywhere." (R11, female, 28)
	• Healthcare reimbursement 9. "What drives me crazy is all the administration I have to deal with. It is one big mess. I think I can't cope with it anymore." (R20, female, 54) 10. "It would be very nice if, when you're faced with such news [ALS diagnosis], you'd get a mentor, who says 'What do you need?' and 'These are the steps you have to go through.'" (R9, female, 35)	
Counseling	• Expressing thoughts and feelings to an independent person 11. "Getting things off your chest, I like that. And I notice I tend to hide my grief. It's in those conversations that it comes out." (R8, female, 64) 12. "What I needed was to talk to someone whom I didn't know, to whom I could tell my story, without having to feel guilty towards my husband." (R19, female, 67)	
	• Accepting diagnosis/dealing with emotions 13. "I feel powerless. Then I think why did he get ALS? Why? There's no answer." (R18, female, 67) 14. "The deterioration [of the patient's condition] simply overcomes you. You just don't have the time or the space to process that your father is ill. (...) It's like getting on a speeding train." (R9, female, 25)	
	• Setting boundaries with regard to care 15. "I'm not assertive enough to set boundaries (...) but partly also because I think 'He's ill, he's dying! Should I say no all the time?' I know that I don't take enough care of myself (...). My time will come, so I'll leave it as it is. But in the meantime, it's destroying me." (R12, female, 29)	
	• Communication with patient 16. "That we [patient, caregiver and counselor] can talk about the end of life. That [talking about the end of life] is also difficult for her. That's something that you keep postponing." (R4, male, 73)	
	• Dealing with patient's behavior problems 17. "I'd like to receive tips on how I can deal with the situation [patient's apathy]. Do I need to push him? Or just leave things as they are?" (R6, female, 38)	
	Peer contact	• Sharing experiences 18. "I would like to speak to someone who had also lost their mum or dad to ALS and, like me, is struggling with it. (...) Because I notice that in my environment, people just can't imagine what it's like." (R9, female, 25)
	• Receiving information and tips 19. "An evening during which people can ask about problems others have encountered and how these problems were solved. I might hear things that I can use." (R11, female, 28)	

help them cope with the diagnosis and their grief because they were struggling with acceptance and feelings of disbelief, anger, injustice, anxiety, sadness, and guilt (quote 13). In some cases, a rapid progression of ALS complicated the situation for caregivers because they felt they did not have enough time to come to terms with the disease (quote 14).

Caregivers expressed the need for support in setting boundaries with regard to their personal needs and the amount and type

of care they provided. Some participants indicated that setting boundaries caused feelings of guilt because it meant they had to express their own needs in the face of someone suffering from a terminal illness (quote 15). Looking back, burdened participants reflected that they had crossed their own boundaries without realizing it.

Some caregivers indicated that it was difficult to share their feelings with the patient or to discuss sensitive topics, such as

death and euthanasia. They wanted support in addressing these topics and enhancing communication (quote 16). Communication was particularly difficult when the person with ALS was unable to speak and had to make use of augmentative and alternative communication. Problems in communication were also provoked by the fact that some patients with ALS had become more withdrawn and less communicative since the diagnosis.

Caregivers noted various degrees of behavioral change in ALS patients since the onset of the disease as a result of the psychological impact of ALS or as a consequence of cognitive problems caused by the disease. Some caregivers indicated they wanted support to help them cope with these behavioral changes. Two behaviors that stood out as being particularly problematic were apathy and demanding behavior. Apathetic patients behaved as if their life had already ended and would not attempt any activities, rarely expressed themselves, and made little effort to make decisions. In these cases, caregivers were burdened with taking over decision-making, for example, with regard to care. Caregivers would like to be advised on ways to cope with apathy in patients (quote 17). Demanding behavior constituted another difficult change because caregivers felt that their family member with ALS made demands that were inconsiderate of others' needs. This led to participants struggling with feelings of not being in charge of their own life and of not being appreciated. Some participants mentioned that it was difficult to justify to themselves that they would like counseling support, worrying that this might be interpreted as an act of egocentrism.

Peer contact

Almost half the caregivers indicated that they would like to talk to other caregivers and share experiences because these people would understand what they were going through in a way that their own social network could not (quote 18). However, only a few participants actively searched for and contacted other caregivers. They did not know how to get in contact with them. Some were hesitant about such contacts, as they felt each situation was different and being confronted with patients in a more progressed state frightened them.

Receiving tips, information, and advice was considered another major benefit of peer support. Caregivers expected that they could receive information from peers about how and when they should apply for care provision, which problems they might encounter, and how they might solve them (quote 19). Some searched the internet (e.g., blogs of other caregivers) to get information about others' experiences and about intimate topics that were difficult to discuss, such as having sex with a severely disabled ALS patient, but this was hard to find, and more information would be welcome. Some would be interested in joining a forum bringing together caregivers of ALS patients to answer each other's questions and share information; however, the majority indicated that they would be passive partakers and would only read the information.

Discussion

ALS caregivers reported a need for more personal time, assistance in applying for resources, counseling, and peer contact. Despite these needs, caregivers were reluctant to apply for and accept support. Balancing their needs with their care responsibilities and the patients' needs was difficult for caregivers, and their priority were the needs and wellbeing of the patient.

Meeting the need for more personal time seems challenging for caregivers of ALS patients, as has also been recognized in other studies in ALS (Ozanne et al., 2012). Caregivers reported that when they used paid home-care, they were confronted with reduced privacy. Additionally, caregivers experienced difficulties in handing over care to others, which often caused distress rather than relief. Handing over care tasks might be difficult in ALS care because of the complexity of the tasks and the lack of experience with these tasks of others who are offering help (Foley et al., 2012). However, these difficulties might also be partly attributed to the caregivers' own perceptions of their caregiving role and the expectations that patients have about care provision (Lerum et al., 2016). This appraisal process might lead to rejection of support and may result in caregivers feeling they have to be continuously available, which results in feelings of burden (Gauthier et al., 2007). Research among dementia caregivers shows that caregivers often wait to seek help until they can no longer cope (Robinson et al., 2005).

Another stress-provoking issue for caregivers is the process of applying for resources, which is generally time-consuming and bureaucratic, whereas the need is often urgent. Unfortunately, caregivers and patients are not able to influence the speed and handling of this application process. Previous research found that the total number of perceived problems with health and social care services has a negative impact on the ALS caregivers' quality of life and strain (Peters et al., 2012). Perceived problems of the application process may lead to a lack of balance between caregiving demands and feelings of control over caregiving tasks and subsequently may lead to distress (Molloy et al., 2008).

Furthermore, caregivers reported a need for counseling on specific topics: setting boundaries, dealing with emotions and acceptance, dealing with behavior problems and communication. Setting boundaries with regard to care often leads to feelings of guilt (Weisser et al., 2015) but seems to be crucial for the wellbeing of the caregiver, especially in the context of caring for a patient with a progressive disease. Interventions for caregivers that focus on topics such as information, dealing with emotions, coping skills, and communication have proven to be effective among caregivers of patients with cancer and might be beneficial for ALS caregivers as well (Waldron et al., 2013). In addition, psycho-education about the patient's cognitive and behavioral decline might help the caregiver deal with behavior problems (Merrilees et al., 2010). Despite these specific needs of caregivers, psychological support for caregivers is not yet a standardized part of ALS care in every multidisciplinary ALS care team (Van den Berg et al., 2004).

Another need among Dutch caregivers was reported to be peer support, having contact with other caregivers to share experiences and exchange helpful advice and information. This would be another potential target for intervention. Although some of the ALS care teams in the Netherlands organize peer support meetings for caregivers, caregivers find it difficult to attend these meetings, due to lack of time and the difficulties of handing over care to others. An online platform may enhance the accessibility and use of peer support groups (Locock et al., 2010).

Contrary to other studies (Bergin et al., 2016; Bruletti et al., 2015), Dutch caregivers in our study did not express a need for information about ALS or training in nursing skills (Bergin et al., 2016; O'Brien et al., 2012). This might be a result of cultural differences and differences in the organization of care (Bede et al., 2011). In the Netherlands, specialized nursing care is predominantly provided by home-care professionals, which might be a

reason why no need for training was reported by the Dutch caregivers. In addition, Internet access in the Netherlands is high and the ALS center has an informative website, which may have resulted in the fact that Dutch caregivers were satisfied with the information provided.

Limitations

Our results need to be viewed in the context of the Dutch ALS care setting, in which specialized ALS care teams provide multi-disciplinary care according to international guidelines (Van den Berg *et al.*, 2005). This may limit the generalizability of our findings.

Implications

Suboptimal support for caregivers may have serious adverse consequences for the patients' wellbeing. Because ALS caregivers report being reluctant to ask for help despite their feelings of burden (Brulletti *et al.*, 2015), a proactive and tailored approach is needed. Many factors can facilitate or hinder the use of a support service, such as the personal characteristics of the caregiver (e.g., perceived need, coping style), relational factors (e.g., relationship with the patient, support from the community), and the characteristics of the service itself (e.g., availability, quality, accessibility) (Mast, 2013). Therefore, it seems essential for the healthcare professionals involved to acknowledge the importance of caregivers in the care of ALS patients at an early stage; inform caregivers about the risk of burden; monitor their wellbeing; and repeatedly offer support opportunities (e.g., support organizations, respite care, psychosocial support). Because caregivers expressed the need for support but also reported lack of time, e-health can be an option to provide support at home in a less time-consuming manner. The majority of caregivers are known to use devices such as tablet computers or laptops on a daily basis and caregivers are open to technology-assisted care (Hobson *et al.*, 2017). E-health may enable online psychological support, online contacts with peers and online information about applying for aids (Wasilewski *et al.*, 2017).

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Conflicts of interest. None.

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