

Balancing needs as a family caregiver in Huntington's disease: a qualitative interview study

Merete Røthing RN MSc^{1,2}, Kirsti Malterud MD PhD^{2,3,4} and Jan C. Frich MD PhD^{5,6}

¹Research Network on Integrated Health Care in Western Norway, Helse Fonna Local Health Authority, Haugesund, Norway, ²Department of Global Public Health and Primary Care, University of Bergen, Norway, ³Research Unit for General Practice, Uni Research Health, Bergen, Norway, ⁴Research Unit for General Practice in Copenhagen, Denmark, ⁵Institute of Health and Society, University of Oslo, Norway and ⁶Department of Neurology, Oslo University Hospital, Norway

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Correspondence

Merete Røthing
Research Network on Integrated
Health Care in Western Norway
Helse Fonna HF
P.O. Box 2170, N-5504 Haugesund,
Norway
E-mail: merete.rothing@helse-fonna.no

What is known about this topic

- Family members experience role conflicts in families with severe chronic disease.
- Care-giving may be experienced as a burden.
- Caregivers report lack of support in their care-giving process from health professionals, social workers and other family members.

What this paper adds

- Family caregivers of patients with Huntington's disease experience isolation and a life on hold, and do not succeed in keeping a balance, thereby compromising their own needs.
- Health professionals and social workers should support and encourage family caregivers in maintaining some own valued activities.
- Information and support should be tailored with respect to trade-offs that caregivers make in deciding on strategies of coping.

Abstract

Family members in families with severe chronic disease play important roles in care-giving. In families affected by Huntington's disease (HD), caregivers encounter practical and emotional challenges and distress. Enduring caregiver burdens may lead to problems and caregivers are in need of social support and health services to deal with challenges. We wanted to explore coping strategies and behaviour patterns used by family caregivers to care for themselves, while caring for a family member with HD. Participants were recruited from hospitals and community-based healthcare. The sample represents experiences from care-giving in all stages of the disease. We conducted semi-structured interviews with 15 family caregivers in Norway. The transcribed material was analysed by use of systematic text condensation, a method for cross-case thematic analysis of qualitative data. We found that family members used various coping strategies, adjusted to the stage and progression of HD. They tried to regulate information about the disease, balancing considerations for protection and disclosure, within and outside the family. The participants made efforts to maintain a balance between their own needs in everyday life and the need for care for affected family member(s). As the disease progressed, the balance was skewed, and the family caregivers' participation in social activities gradually decreased, resulting in experiences of isolation and frustration. In later stages of the disease, the need for care gradually overshadowed the caregivers' own activities, and they put their own life on hold. Health professionals and social workers should acknowledge that family caregivers balance their needs and considerations in coping with HD. They should, therefore, tailor healthcare services and social support to family caregivers' needs during the different stages of HD to improve caregivers' abilities to maintain some of their own activities, in balance with care-giving.

Keywords: caregivers, chronic diseases, coping, Huntington's disease, support

Introduction

Family members play an important part in providing care and take on different roles as caregivers for patients with chronic diseases (Nolan 2001, Fisher & Weihs 2000, Gillick 2013). Severe chronic disease often causes a functional decline in the affected person, and the family caregiver, as well

as the family, may encounter practical and emotional challenges and burdens over longer periods of time (Bastawrous 2013). Health services are increasingly considering family caregivers to be partners in collaborative teams, to achieve higher quality and effectiveness in the care-giving process (Collins & Swartz 2011, Fisher & Weihs 2000, Lilly *et al.* 2012). Family carers are in need of support which enables them to cope with the challenges, to take care of themselves and stay healthy to endure the care-giving course (Nolan 2001, Acton 2002, Collins & Swartz 2011).

Huntington's disease (HD) is a severe chronic hereditary and neurodegenerative disease, characterised by cognitive decline, motor disturbances and psychiatric symptoms. Gradually, the affected person's ability to function in everyday life decreases. The average age of onset is 40 years, but psychiatric symptoms and changes in cognitive functions are often reported prior to the clinical diagnosis (Novak & Tabrizi 2010). HD progresses over five stages, with a disease duration of 15–20 years, and the progression and severity of the symptoms may vary between individuals. In the early and mid-stages, the affected person usually lives at home with assistance from family members or professionals from community-based health services. In later stages, most patients are in need of institutional care (Novak & Tabrizi 2010).

Previous research has revealed how caregivers are burdened in their role as a caregiver. Family members experience care-giving in HD as burdensome and stressful, and the experience has been characterised as a 'lonely ride' (Etchegary 2011, Soares 2012, Williams *et al.* 2012). Living with HD in the family may also cause role conflicts and strain among family members (Røthing *et al.* 2013), and caregivers may face and deal with practical and emotional distress (Roscoe *et al.* 2009, Aubeeluck *et al.* 2012). As HD is a rare disease, health professionals and social workers in general are not well experienced in working with individuals and families affected by HD.

Lack of HD knowledge and experience among community health professionals might complicate collaboration possibilities of understanding caregivers' challenges and needs, and lack of support from other family members increases caregiver burdens (Helder *et al.* 2002, Dawson *et al.* 2004). Caregivers use a variety of coping strategies, but further research is needed to investigate how caregivers might be supported in their coping in the specific situation as caregiver in families with HD (Helder *et al.* 2002, Soltysiak *et al.* 2008).

Stress has been conceptualised in different ways, but there seems to be agreement that stress is a stim-

ulus–response transaction based on how an individual perceives a stressor (Weiten *et al.* 2012). Lazarus and Folkman (1984) suggest that stress is determined by an individual's considerations of the possibilities for coping, and stress thus becomes a question of a balance between resources and demands. Within this stress model, 'transaction' refers to the mutual and dynamic relationships between a person and circumstances of the environment where stressful events take place. Chronic disease may represent a stressor, and the context, social support and an individual's personal resources may determine the experience of stress (Weiten *et al.* 2012). Coping strategies can be categorised into problem-focused, appraisal-focused and emotional-focused strategies. *Problem-focused* strategies aim at changing or alleviating the source of stress; *appraisal-focused* strategies may be used when there is no easy solution, and people cope by changing the way they think; while *emotional-orientated* coping strategies involve dealing with emotions caused by stress. The use of constructive strategies is based on an acceptance of a problem, but is not necessarily promising success.

Knowledge about family caregivers' experiences of coping during the course of HD may enable professionals to understand caregiver needs, and facilitate collaboration during the course of the care-giving process. The authors of this article have professional backgrounds from nursing and medicine; have experience in specialised neurology hospital wards, community healthcare and general practice. We are familiar with some of the challenges family caregivers face, specifically, with the characteristics of HD. In this study, our aim was to explore coping strategies and behaviour patterns used by family caregivers to care for themselves, while caring for a family member with HD.

Methods

Design and sample

We conducted a qualitative study based on individual, semi-structured interviews, as we wanted to explore variations in experiences of coping and care-giving from the perspective of family members in care-giving roles (Polit & Beck 2004, Pope & Mays 2006). Our sample consisted of 15 participants recruited through a written request sent from hospital departments, local health authority in communities responsible for nursing and follow-up programs for persons affected by chronic disease and a lay organisation for HD in Norway. The Regional Committee for Medical and Health Research Ethics

approved the study prior to data collection (REC South-East B, ref. 2010/2072). All the participants received an information letter about the purpose of the study. They were informed that participation was voluntary and that they could withdraw from the study without any consequences for the services they received.

The participants' ages ranged from 20 to 67 years, with three males and 12 females. The sample consisted of individuals with experiences in caring for an HD-affected parent, spouse, siblings or adult children. The average care-giving experience was 11.6 years, and the experiences represented all five stages of HD. With regard to the main occupations outside the home, the participants worked full- or part-time, or were students (see Table 1 for further details).

Data collection

Data were developed from semi-structured individual interviews (Pope & Mays 2006). The interviews were conducted by the first author from October 2011 until February 2012. The participants chose where and when the interviews would be conducted. Some took place in their home; others preferred an office in a local health institution. Each interview lasted 60–90 minutes. The affected person was not invited, and for several of the participants, it was difficult to make an appointment fit into their care-giving schedule. The present study is part of a larger project and the

interviews were based on a thematic interview guide with three different themes: participants' experiences as a family caregiver, descriptions of how they dealt with behavioural challenges and what coping strategies they used, and their experiences with the health services. In this article, we focus on the findings related to coping strategies. The interview guide was developed with inputs from HD specialists in a hospital department and a group of three family caregivers. Emerging themes and hypotheses were explored in the interviews with subsequent participants. The material was digitally recorded and transcribed verbatim by the first author.

Data analysis

In the data analyses, we used systematic text condensation, a method for the cross-case thematic analysis of qualitative data (Malterud 2012), as follows: (i) reading all of the material to obtain an overall impression of family caregivers' coping strategies and experiences; (ii) identifying meaning units, representing aspects of coping strategies applied by participants to live with HD in the family, and coding for these; (iii) condensing the contents of code groups and subgroups; and (iv) summarising the meaning from each code group to generalise descriptions and concepts concerning coping strategies and experiences. All authors read the material, informed by theories of coping. In the first step of analysis, 'taking care of oneself' emerged as a substantial theme, as well as the importance of maintaining a social life outside the role as caregiver. These themes were then further elaborated into code groups and subgroups (Malterud 2012). The authors discussed and interpreted the data, identifying strategies and the various aspects of how caregivers handled behaviour as individuals and within the family.

Findings

Our analysis demonstrated a variety in coping strategies used by family members, adjusted to the stage and progression of the disease. They tried to regulate information about HD, balancing considerations for protection and disclosure, within and outside the family. The participants made efforts to balance their own needs for activities in their everyday lives, and the need to care for an affected family member. In later stages of the disease, the need for care gradually overshadowed the family caregivers' own activities, and they put their lives on hold. We elaborate further on these findings below.

Table 1 Characteristics of 15 family caregivers

	N (%)
Age in years	
20–29	1 (7)
30–39	0 (0)
40–49	6 (40)
50–59	4 (27)
≥60	4 (27)
Gender	
Female	12 (80)
Male	3 (20)
Family caregiver's position	
Spouse	10 (67)
Ex-spouse	1 (7)
Child of affected individual	4 (27)
Caring for multiple family members	
Yes	3 (20)
No	12 (80)
Family caregivers have children	
Yes	12 (80)
No	3 (20)

Regulating information about the disease

The caregivers responded that they obtained knowledge about the disease from healthcare professionals, family members or the internet, which represented a resource to help understand why their affected family member had changed their behaviour and personality. Information about HD could, however, also cause concerns for children and grandchildren at risk, and the regulation of information became an important issue for caregivers. They described how they regulated information to protect themselves or the affected family member against the knowledge of possible future challenges and also as a protection from negative reactions of others. Participants told for example that they would hide literature and pictures of possible problematic conditions of the disease trajectory or care equipment from family members. Caregivers said that they withheld knowledge about heredity from their children to avoid stress, and mothers of children at risk explained that they had held back information from their teens about heredity because they wanted them to have normal 'puppy love'. Regulating information involved a balance between protection and disclosure, as illustrated by a caregiver who had chosen to be open about HD with her employer, but was asked by her husband not to share the information with his family:

I have chosen to be open about it at work, but not with my affected husband's family. His daughter's parents-in-law can see that something is wrong, but we are not allowed to talk about the reason why. His daughter has been tested, and has her own children. They want to protect her husband's family from knowing about HD. (E2)

Caregivers said that they were open about HD when they considered it appropriate, such as informing their children's teachers, hoping to improve understanding and support in case there were reactions at school. To enjoy friendship and social participation on their own premises, the participants revealed that they had to control when and with whom they shared information. As the disease progressed, the affected person's functional decline became more visible. To avoid rumours and speculations, participants chose to be more open about the diagnosis than they were in the earlier stages. However, they often chose not to disclose that the illness was hereditary. Some caregivers expressed positive experiences with such strategic openness in the early stages of HD. They could deliberately make a decision to inform their children, as illustrated in this quote by the spouse of an affected husband. She had earlier positive experiences from collaborations with

healthcare professionals related to other conditions concerning one of their children:

I thought it was ok that people around here know about us – so they can take our situation into account. I thought it was better that our children heard about the disease from their mother [...] about what was going on with their dad, the rumours around here, about him sitting in a wheelchair. (B3)

Towards a skewed balance and increasing isolation

The caregivers explained how maintaining a normal, everyday life, such as socialising with friends or colleagues without focusing on the problems, was valuable. In the early stages of the disease, changes in behaviour and a decline in the function of an affected family member had social consequences, primarily for the affected person's participation in everyday life activities. Participants reported experiencing emotional difficulties such as anxiety, but seemed to have adjusted to minor care-giving tasks and changes. While some adaptation was necessary in the early stages, there was the possibility for caregivers to maintain their daily activities. A woman, whose husband had his HD onset in his late 50s, described how she and her family had adapted to the changes in her everyday activities:

My husband is not able to work anymore because of the disease, but our son is now in charge of our little business and I can still work as before, three days a week. We have just reorganised a little bit. He helps me in the house, and I still keep in touch with my friends and see them regularly. (B1)

As HD progressed, caregivers met new demands that challenged their previous strategies to maintain their own lives. The participants described how they gradually became aware of the increased care burden. Male as well as female participants had considered solving the problem by leaving the affected spouse or partner. One participant decided to divorce and moved out with the children, as she considered a role as main family caregiver hard to combine with the need for care of their young children. A man whose wife was affected by HD had considered a divorce, but the situation of the illness made him refrain from it:

She was difficult to live with for a period. If she had been healthy I would have left her, but then I understood she was very ill. (A2)

The caregivers described their sense of responsibility, feelings of guilt and pity for their family members' destinies. They explained that while they earlier had balanced care-giving and maintained their own lives, this was no longer an adequate strategy. While

trying to adapt to new care challenges, caregivers gradually lost their attention and resources in relation to their own needs, and the balance was gradually skewed. They had to reduce their own social activities which resulted in less social contact and increased isolation. Leaving home for activities, knowing that the affected family member had been alone all day, could be difficult. Still, caregivers articulated a need for respite, described as breaks from caring responsibilities and tasks in the family. Respite, they said, represented space for expressing their own feelings and fulfilling their own needs, such as a vacation without caring obligations. A break could be time for themselves, relaxation or a diversion from care-giving. One of the eldest participants said that to him, respite was a weekend trip with his grandchildren. However, the participants revealed that possibilities for breaks were gradually reduced for practical reasons. The caregivers would gradually reduce their leisure activities, such as outdoor activities or spontaneous gatherings with friends. A female spouse who worked full-time said:

Earlier I could join a running group, we were jogging and talked together, but now I am almost always at home with him [...] on the weekend we try to do things together [...] I feel bad about that. He likes to be outside, but I have to be with him all the time. He occupies my time. (B5)

Increasing limitations – tolerating a life on hold

The caregivers described an everyday life in the late stages of the disease as living in the shadow of HD. During the progression of the disease, they had gradually given up most of their activities, and important aspects of their own lives were put aside. The participants still tried to maintain their role as employees. Work outside the home was appreciated, because it offered social contact with colleagues, value as individuals when involved in their work and financial independence:

I have chosen to work full-time, actually now also with the possibility of working at home, to reduce overtime work. After an accident, my shoulder was not well and my physiotherapist advised me to stay at home. But, I prefer to go to work because it is better. Somehow, my function at work is better than my function at home; my work keeps me going, with good colleagues. It means a lot to me, it is the only place I can socialise. (B2)

One female participant described how work outside the home represented the possibility of an 'HD-free zone' that was important for her health, although the boundaries between her work life and life at

home could be difficult to maintain. Several participants explained how an affected spouse, due to the decline in cognitive functions, disturbed them at work with unwanted visits and telephone calls several times a day. One participant said she kept memories of a previously stable and happy family life with her husband as a good father for their children. A dignified caring process was important for her. She had given up work outside the home:

His situation of HD has occupied my life, as an elephant. There is no space left for anything else. He is looming in my life. [...] I have made a choice, I continuously assess my situation and the needs of our children, and meanwhile I put my life on hold. I am not bitter, and have managed to find occasions for breaks, knowing he is taken well care of. (E4)

Several participants had received invitations to educational programmes for patients and caregivers. While some had participated in those programs, others declined because they were afraid of being emotionally involved in another caregiver's situation. They were also afraid of not being understood. 'Take care of yourself' was the advice several of the participants had received from friends and healthcare professionals when they described their situation. How one could take care of oneself was, however, seldom mentioned. One of the family caregivers described how she took care of herself by living a limited and regular life:

I try to stay healthy. Sleep as much as I can and go for walks alone. I live a regulated life. Sometimes, it happens, I find time for a little trip, alone or with my children. It is rare. I need something to look forward to and something nice to think of later. I have to, if not, I know I will be depressed. (B5)

One group of participants had experiences from care-giving as children and teenagers, and they said that they gradually cared more for the affected parent's functions in their family, while their own needs were put aside. They had experiences of not being able to bring home friends or have time to stay outside in the afternoon, and homework and school were given low priorities. In the late phase of HD, after years of care-giving but still being adolescents, they revealed that care-giving overshadowed their own needs and that their possibilities for living their own lives were minimal. To establish lives of their own, the adolescents therefore found it necessary to leave home:

I knew that if I should manage to take care of myself, I had to start thinking about my own life, and could not manage to take care of my father and siblings. It was simple; I was empty, nothing more to give to them. (E1)

Discussion

The family caregivers of patients with HD cope by regulating information about the disease, and balancing their own needs with the need to care for a loved one. Gradually, they experience isolation and a life on hold, and do not succeed in keeping a balance, thereby compromising their own needs. Below, we discuss the strengths and limitations of these findings in the context of existing research within this field.

Acceptance and sense making

Although the family caretakers in our study seemed to accept the HD diagnosis, they were very selective in talking to others about their situations. In the early stages of the disease, they continued with social participation and work. This is consistent with findings from previous research, where spouses of people affected by HD scored highly on acceptance (Helder *et al.* 2002). The more seriously the spouses perceived the symptoms and the duration of the illness, the less they were inclined to use denial strategies. Another study showed that spouses of HD-affected family members avoided talking about the disease in general, often by denial (Lowit & Van Teijlingen 2005). Our study adds to existing knowledge by describing how participants in later stages of HD still seemed to accept the diagnosis, although a balance, as in earlier stages, was more difficult to maintain. They were frustrated and did not see possible strategies to solve the problem and re-establish balance, an appraisal-focused strategy, related to new experiences of emotionally orientated challenges (Lazarus & Folkman 1984). Similarities in caregiver experiences of emotional impacts have been documented across other more common progressive diseases (Figueiredo *et al.* 2014, Grose *et al.* 2013). Symptoms of HD and questions of inheritance are of such a nature that particular attention should be given to the caregiver's individual needs for support in different stages of the care-giving course.

Managing information

The participants regulated how much and which information about HD they shared with their family members, friends and colleagues. We are not the first to describe how fear of characteristic symptoms and stigmatisation of families affected by HD is part of HD history, such as holding back information about an affected grandparent or speaking of early death of a parent (Wexler 2010). Our study contributes in particular with descriptions of how participants used

both emotion-focused and problem-focused coping strategies. Use of a problem-focused strategy in one situation could facilitate or complicate the use of an emotion-focused strategy in another. For example, while openness about HD might have solved the problem of others speculating about the cause of the symptoms, and mobilised practical support in an early stage, openness may lead to emotional challenges related to genetic questions. The two different strategies may be seen as interrelated, sometimes complementary, but having different functions in different stages of HD (Lazarus & Folkman 1984).

Carver and Connor-Smith (2010) discuss proactive coping as part of a problem-focused strategy, intended to prevent possible expected stressful situations from arising. The problem-coping behaviour of the participants in our study may be a strategy to manage a problem that has arisen as the result of an emotion-focused strategy. Family caregivers in our study, with experiences from care-giving as a child or adolescent, seemed to use coping strategies similar to adults. They did not have the life experiences and social relationships to assess their situation, which could cause distress. Information early in life might be of importance. Previous research shows that young persons who had been given information, and grew up knowing about HD from an early age, seemed to cope better later in life (McCabe & O'Connor 2012). Progression and severity of symptoms may vary, as does the time needed for caregivers to adjust to changes and to be prepared for new challenges in later stages. Informing family members about the diagnosis and possible progression seems not to be a one-time task in an early stage. Our findings indicate that there is a continuous need for information and support from healthcare professionals, tailored to the specific situation, especially concerning young caregivers.

Maintaining a life of one's own

We found that family caregivers experienced increasing difficulties in maintaining their own social lives as HD progressed. HD families struggle with increasing isolation and complicated family relationships among extended family members (Lowit & Van Teijlingen 2005, Soares 2012). Caregivers for persons with HD experience the greatest loss of social relationships, compared with caregivers of persons with different diagnoses, such as motor neurone disease, multiple sclerosis and Parkinson's disease (McCabe *et al.* 2009). Social support from family members and other individuals is highlighted as an important factor for successful adjustment to chronic illness.

It seems as if participants in our study reached a threshold where they no longer could maintain valued activities because the care-giving tasks at hand exceeded the available resources. Our findings add to previous knowledge by illustrating the impact of changes in social context and mutuality between an individual and environment for coping (Lazarus & Folkman 1984). Our findings that family caregivers' work compensated for some of the losses due to decreased social activity are supported by previous studies (McCabe *et al.* 2008). Furthermore, our analysis suggests that attention to the gradual changes might be essential for possible early interventions to prevent isolation. To maintain a social life throughout the care-giving process, adequate social support is necessary. Our findings emphasise the need for tailored support and assistance. Family caregivers explained that participating in social activities and work was essential for their well-being, contributing to physical health, value and recreation. We found that these needs were compromised during the course of HD. Previous research, also related to severe diseases other than HD, suggests that less physical activity, disturbed sleep or lack of rest could impact the family caregiver's quality of life in a negative way over time (Aubeeluck *et al.* 2012, Northouse *et al.* 2012, Lawang *et al.* 2013). Apart from the questions of inheritance and possibilities for treatment, care-giving in HD might have a similar influence on the family members' own lives and well-being.

Methodological considerations

Semi-structured individual interviews offered the possibility to follow individual trajectories of sensitive experiences, without having to consider other individuals, although focus group interviews might have opened up the sharing of additional experiences (Pope & Mays 2006). Coping was a central theoretical term in our study. Our interview guide was developed with inputs from experienced family caregivers who emphasised the importance of wording in the interview situation. They advised us to use colloquial phrases for coping behaviour, such as 'How do you handle this?' and 'What do you do?' In the interview situation, the interviewer tried to ask for descriptions and concrete examples of their efforts. The diversity of the participants' contributions has probably been strengthened by the details of the wording.

Three researchers with different backgrounds as healthcare professionals were involved in interpreting the data, which we consider to be a methodological strength. However, a family therapist may have noticed other aspects of social interaction or commu-

nication in the family and focused differently in follow-up discussions (Malterud 2001). There are certainly HD caregiver experiences that our design did not illuminate. Although we included all the men who responded, we have limited information on young male family members' experiences. Some participants were interviewed while being faced with complicated and emotionally challenging decisions concerning the needs for palliative care and ending home-based care. Their accounts of care-giving over the years were shaped by their present situation. Still, we were able to discover that coping strategies seemed to change during the course of the disease.

We consider our results to be transferable to family caregivers in families affected by HD, while still recognising that extended family members may have different roles in other cultures, perhaps more involvement in care-giving. HD is a rare disease, with characteristics unlike other more common progressive diseases. Personal and cultural values, individual priorities and the stage in the life-cycle all impact the care-giving process. Taking this into account, we consider some of our findings about the value of maintaining activities to be transferable to care-giving processes in other severe chronic diseases.

Implications for practice

Our findings indicate that health professionals and social workers should support and encourage family caregivers to maintain some own valued activities and offer flexible services. Our analysis suggests that information and education programs should be oriented towards changes in caregivers' lives, and information and support should be tailored with respect to trade-offs that caregivers make in deciding on strategies of coping. Professionals should be aware of the risk of caregivers being isolated from family members, relatives or social participation. Our findings also indicate that a wide approach to adequate resources might be useful, including collaboration with employers.

Conclusions

Health professionals and social workers should acknowledge that family caregivers balance needs and considerations in coping with HD. They should tailor information and support to family caregivers' needs in different stages of HD.

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Contributions

Study design: MR and JCF; Data collection and analysis: MR, KM and JCF; Manuscript writing: MR, KM and JCF.

Conflict of interest

There is no conflict of interests.

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