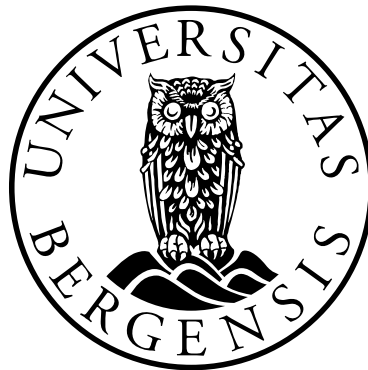


# **Towards improved partnerships between health professionals and family caregivers in Huntington's disease: a qualitative study**

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## **SCIENTIFIC ENVIRONMENT**

This thesis has been accomplished within the institutional framework of the doctorate program, Faculty of Medicine and Dentistry, University of Bergen.

The research work, courses and co-supervision are institutionally affiliated with the Research Group for General Practice (ALFO), Department of Global Public Health and Primary Care, University of Bergen. Jan C. Frich at the University of Oslo and Kirsti Malterud at the University of Bergen have supervised the research work on this thesis. In my daily work with this project, I have been affiliated with Research Network on Integrated Health Care, in Western Norway, Helse Fonna Local Health Authority in Haugesund.

## PREFACE

It has been a privilege to conduct this research, to which many have contributed with great knowledge and support. With all my respect, I am grateful to the participants in this study for sharing their stories, concerns and hopes with me.

I offer many thanks to my main supervisor, Jan C Frich. You have been involved from the beginning and have been a pillar throughout the process. You have shared your knowledge and wisdom and have patiently guided me through the difficult seasons in this process. I also thank my co-supervisor, Kirsti Malterud. You have inspired my work with your standards of excellence and your knowledge, and especially your fearlessness to question methods and theoretical perspectives. Together, you two have been the powerhouse behind my work, always willing to find new solutions for improvement in the research process.

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## LIST OF PAPERS

### **Paper I**

Røthing M, Malterud K, Frich JC. Caregiver roles in families affected by Huntington's disease: a qualitative interview study. *Scand J Caring Sci.* 2014; 28(4):700-5.

### **Paper II**

Røthing M, Malterud K, Frich JC. Balancing needs as a family caregiver in Huntington's disease: a qualitative interview study. *Health Soc Care Community.* 2015; 23(5):569-76.

### **Paper III**

Røthing M, Malterud K, Frich JC. Family caregivers' views on coordination of care in Huntington's disease: a qualitative study. *Scand J Caring Sci.* 2015; doi: 10.1111/scs.12212. [Epub ahead of print]

I will refer to the papers by their Roman numerals.

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## ABBREVIATIONS

CAG	Codon for the amino acid glutamine
CFAM	Calgary Family Assessment Model
CHDI	Cure Huntington's Disease Initiative Foundation
EHDN	European Huntington's Disease Network
FST	Family Systems Theory
GP	General Practitioner
GST	General System Theory
HD	Huntington's Disease
HDQoL-C	Huntington's Disease Quality of Life Battery for Carers
HTT	Huntingtin, a gene protein
IP	Individual Care Plan
RC	Relational Coordination
STC	Systematic Text Condensation
TFC	Total Functional Capacity Scale

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## ABSTRACT

**Background:** Huntington's disease (HD) is an autosomal-dominant inherited neurological disease characterised by progressive cognitive, motoric and behavioural symptoms. HD results in loss of functions and an increasing need for health care services in the home and later in a nursing home and there is still no cure. There is a need for better understanding of the impact of HD on families and family caregivers and ways to develop constructive collaboration between families and health professionals.

**Aims of the study:** The overall aim of this study is to contribute with knowledge to promote and support partnerships between health professionals and family caregivers managing HD. This thesis is based on three sub-studies with three specific aims. These were to explore 1) family caregivers' experiences with the impact of HD on the family structure, dynamics and roles in the family, 2) coping strategies and behaviour patterns used by family caregivers to care for themselves while caring for a family member with HD, and 3) experiences and expectations of family caregivers' concerning collaboration with health professionals.

**Methods:** We invited family caregivers to participate by request through hospital departments, health institutions in municipalities and the Norwegian Huntington Association. In a qualitative study, we conducted individual semi-structured interviews with 15 adult participants in caregiver roles. Data were analysed with systematic text condensation (STC).

**Results:** Our results suggest that the family caregiver role is shaped by gradual changes in family dynamics as decreased functioning of the HD-affected family member and the corresponding needs for care develop. Family caregivers reported conflicts between the roles of caregiver and individual family member (a role wherein they had needs of their own). This was particularly evident when family caregivers described experiences as caregivers in childhood or young adolescence, burdened with care responsibilities normally provided by adult family members. Family caregivers

experienced changes in the qualities of familiar relationships. Emotional challenges related to HD characteristics and family members' different reactions to the illness rendered families vulnerable to fragmentation and lack of social support. In the early stages of the disease, family caregivers searched for information about HD and wanted to establish trustful relationships with health professionals. To manage challenges in the illness situation, family caregivers tried to balance their own needs for daily activities and the family members' needs for care. In the later stages, family caregivers experienced HD as overshadowing other activities of everyday life and felt that their own lives were “on hold”. Participants wanted to take part in daily work outside the home because they felt valued as individuals and needed the social support. Family caregivers expected to be involved in collaboration with health professionals and desired acknowledgment for their competence in providing HD care in the home setting and changes in the family. Participants expected to encounter health professionals with knowledge of HD, and time to maintain trustful relationships and continuous knowledge sharing. However, in their encounters with health care services, family caregivers reported lack of coordination of care services and unclear roles and responsibilities among involved parties.

**Conclusions:** Health professionals should assess family needs and must acknowledge the competence of family caregivers in the context of home care. The family caregiver role is important to recognise through establishment of partnerships based on shared knowledge, adjusted individual support and flexible health care. It is necessary to give children and young family members special attention related to needs and support. Health professionals should arrange for adjusted support that allows caregivers to combine caregiving with valued personal activities, and prevent family fragmentation. To improve the quality of care, health professionals should establish clarity of roles and distribution of responsibilities in a coordinated care course that includes the preferences of family caregivers.



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## SAMMENDRAG

**Bakgrunn:** Huntington's sykdom (HS) er en autosomal dominant arvelig, nevrologisk sykdom karakterisert ved gradvis økende kognitive, motoriske og atferdsmessige symptomer. HS resulterer i funksjonstap med økende behov for helsetjenester i hjemmet og senere i omsorgsinstitusjon. Foreløpig er behandling for HS ukjent. Det er behov for bedre forståelse av hvordan HS påvirker familier og pårørende, og måter å utvikle et fruktbart samarbeid på mellom familier og helsepersonell.

**Mål:** Et overordnet mål for dette arbeidet er å bidra med kunnskap som fremmer samhandling og partnerskap mellom helsepersonell og pårørende i familier med HS. Avhandlingen er basert på tre delstudier som skulle undersøke: 1) hvordan pårørende opplever at HS påvirker familiestrukturen og roller i familien, 2) hvordan pårørende mestrer å ta vare på seg selv og leve med HS, og 3) pårørende sine erfaringer og forventninger gjeldende samarbeid med helsepersonell.

**Metode:** Pårørende ble invitert til deltakelse med forespørsler via avdelinger i sykehus, kommunale helseinstitusjoner og Landsforening for Huntington sykdom i Norge. I en kvalitativ studie gjennomførte vi individuelle semistrukturerte intervju med 15 voksne deltakere med omsorgsoppgaver. Datamaterialet ble analysert med systematisk tekstkondensering (STC).

**Resultater:** Våre resultater viser at rollen som omsorgsgiver formes av gradvise endringer som oppstår i familiemønsteret etter hvert som funksjonstap med tilsvarende omsorgsbehov hos den HS syke utvikler seg. Pårørende opplevde konflikt mellom omsorgsrollen og egne behov som individ og familiemedlem. Det ble særlig tydelig når deltakere beskrev sine tidligere erfaringer som omsorgsgiver i barndommen eller ungdomstiden med oppgaver som normalt tilligger voksne omsorgspersoner i en familie. Pårørende erfarte at kvaliteter i familiære forhold endret seg underveis. Emosjonelle utfordringer knyttet til karakteristika ved HS og familiemedlemmer sine individuelle reaksjoner og opplevelser av tap gjorde familier sårbare for fragmentering og mangel på sosial støtte. I tidlige faser av sykdommen søkte pårørende informasjon

om HS og å få etablert et tillitsfullt kontaktforhold med helsepersonell. For å mestre utfordringer i sykdomsforløpet forsøkte pårørende å balansere egne behov for daglige aktiviteter og behovet for omsorg. I senere faser erfarte pårørende at HS overskygget andre aktiviteter i hverdagen, og deres eget liv ble satt på vent. Pårørende ønsket å delta i arbeidslivet fordi de følte seg verdsatt som individer og fikk sosial støtte. Pårørende hadde forventninger om å bli involvert i samarbeid med helsepersonell og få anerkjennelse for sin kunnskap fra omsorgsarbeidet og hvordan HS påvirket familielivet og familiemedlemmene. Pårørende hadde forventninger til at helsepersonell hadde kunnskap om HS og tid til å etablere tillitsfulle samhandlingsrelasjoner og kontinuerlig kunne dele kunnskap med hverandre. I møte med helsetjenester opplevde pårørende mangel på koordinering, uklare roller og utydelig ansvarsfordeling mellom involverte parter.

**Konklusjoner:** Helsepersonell bør vurdere familiens behov og må anerkjenne pårørende sin kompetanse om konteksten for omsorgsoppgaver. Pårørende sin rolle som omsorgsgiver og samarbeidspartner kan verdsettes gjennom å etablere samarbeidsrelasjoner med vekt på gjensidig kunnskapsutveksling, individuell støtte og fleksible helsetjenester. Barn og unge familiemedlemmers behov for støtte og omsorg må særskilt ivaretas. Helse personell bør også legge til rette for pårørende sin mulighet til å kombinere langvarige omsorgsoppgaver med egne verdifulle aktiviteter og forebygge fragmentering av familien. Det er behov for å øke kvaliteten i helsetjenestene ved at helsevesenet avklarer roller og er tydelige på fordeling av ansvarsoppgaver i et koordinert tjenesteforløp, som også ivaretar pårørende sine preferanser.

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# CONTENTS

SCIENTIFIC ENVIRONMENT .....	I
PREFACE .....	II
LIST OF PAPERS .....	III
ABBREVIATIONS .....	IV
ABSTRACT .....	V
SAMMENDRAG .....	VII
CONTENTS .....	IX
<b>1. BACKGROUND .....</b>	<b>1</b>
PRECONCEPTIONS .....	1
CAREGIVERS .....	2
ESTABLISHING PARTNERSHIPS .....	4
HEALTH CARE IN NORWAY .....	6
HUNTINGTON'S DISEASE .....	9
CAREGIVERS IN HUNTINGTON'S DISEASE .....	12
<b>2. AIMS OF THE STUDY .....</b>	<b>16</b>
<b>3. THEORETICAL PERSPECTIVES .....</b>	<b>17</b>
FAMILY SYSTEMS THEORY .....	17
COPING AND COPING STRATEGIES .....	19
RELATIONAL COORDINATION .....	21
<b>4. DESIGN, SAMPLE AND METHOD .....</b>	<b>25</b>
DESIGN .....	25
SAMPLE .....	25
METHODS .....	27
<i>Data collection</i> .....	27
<i>Role of theoretical perspectives</i> .....	27
<i>Data analysis</i> .....	28
ETHICAL CONSIDERATIONS .....	30
<b>5. MAIN FINDINGS .....</b>	<b>33</b>
PAPER I .....	33
<i>Caregiver roles in families affected by Huntington's disease: a qualitative interview study.</i> ...	33
PAPER II .....	35
<i>Balancing needs as a family caregiver in Huntington's disease: a qualitative interview study.</i> 35	
PAPER III .....	37
<i>Family caregivers' views on coordination of care in Huntington's disease: a qualitative study</i> 37	
<b>6. DISCUSSION .....</b>	<b>39</b>

---

METHODOLOGICAL CONSIDERATIONS .....	39
<i>Reflexivity</i> .....	39
<i>Internal validity</i> .....	42
<i>External validity</i> .....	44
<i>Ethical issues</i> .....	46
DISCUSSION OF FINDINGS.....	47
<i>Coping with the progression of HD</i> .....	48
<i>Special challenges for young family caregivers</i> .....	51
<i>Preconditions for developing partnerships</i> .....	53
<i>Challenges for professional practice</i> .....	56
<b>7. CONCLUSIONS .....</b>	<b>61</b>
<b>8. IMPLICATIONS FOR PRACTICE .....</b>	<b>63</b>
<b>9. FUTURE RESEARCH .....</b>	<b>64</b>
<b>10. REFERENCES .....</b>	<b>65</b>
<b>APPENDIX .....</b>	
DATA ANALYSIS WITH SYSTEMATIC TEXT CONDENSATION.....	
PAPERS.....	
<i>Paper I</i> .....	
<i>Paper II</i> .....	
<i>Paper III</i> .....	

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# 1. BACKGROUND

## Preconceptions

As a nurse, employed as part of a multidisciplinary team in a hospital, I met persons affected by *Huntington's disease* (HD). Occasionally, I had separate consultations with spouses of affected persons. As part of an ambulant work function, I was in a position to visit the affected person at home after hospital discharge, to plan for follow-up programs with community health care professionals. In my encounters with spouses in the home setting, I often found that they presented a different version of the situation than the one given at the hospital. I also found that there seemed to be a gap between their needs for support and the health care provided. I seldom met other family members, such as children, even though parents often referred to them as “needing to talk” with health professionals. My interest in the phenomenon of *family caregivers* originates from these encounters.

Severe chronic disease may trigger specific care needs during a prolonged illness course, and health care delivery may be complex and multidisciplinary. Patients, family members as caregivers and health professionals from specialised medical institutions and community-based primary care may be involved over long periods. Collaboration is especially relevant in HD because the gradual functional decrease of affected person may lead to an enduring need for support from multiple health and social care professionals.

Within the framework of this thesis I considered it too extensive to study all stakeholders' perspectives on challenges and needs being faced with a severe chronic disease. Therefore, I decided to investigate needs and experiences related to caregiving in an HD illness course from the perspective of next of kin as caregivers, focusing challenges of caregiving and collaboration with health professionals, and possibilities to make improvements in health care delivery.

## Caregivers

Collins and Swartz point out that *caregiver* is a broad term. It is associated with specific care responsibilities, care needs beyond normal interactive behaviour due to specific conditions. The caregiver role is not necessarily related to specific personal characteristics or diseases but refers to a person's contribution of care (1). Carduff et al. refer to a definition used in the United Kingdom (UK): “A person of any age, adult or child, who provides unpaid support to a partner, child, relative or friend who couldn't manage to live independently or whose health or wellbeing would deteriorate without this help. This could be due to frailty, disability or serious health condition, mental ill health or substance misuse” (2). *Informal caregiver* is another term used for unpaid family caregivers as opposed to paid, formal health workers. *Professional caregiver* is a term used for health and social workers employed in private or public health contexts, as well as social units reimbursed for services (3).

*Family caregiver* is used broadly as a concept to include partners and other persons who provide or manage care for a person with severe illness. Some authors argue that the term acknowledges the reality of the modern family structure, where most caregiving takes place in the context of family life and a family caregiver is a family member in the role of caregiver (4). Family members are important providers of care, and the main recipients of this care are elderly persons in the last stages of life. There is no specific definition for family caregiving, and the term is used differently in research. Some studies refer to objective measures of caregiving, such as hours of caregiving per day, whereas others rely on the family members' self-identification as caregiver (5).

Caregivers' contributions are becoming highly valued as a health care resource in Western societies. The escalating need for care related to old age and dementia has led to discussions about the role of informal caregivers as a significant part of the total care work force (3, 4, 6, 7). Data from the United States (US) in 2013 suggest that 14% of the US population served as caregivers to adult patients, and that the majority of these caregivers were middle-aged women caring for aging parents (8). An updated

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research report from National Alliance for Caregiving and AARP Public Policy Institute, suggests an increasing estimated prevalence of 16.6% of family caregiving for an adult in US. The majority of US unpaid caregivers are still women, but 40% are male (9). Research related to chronic conditions in general shows that the caregiver role is associated with physical, psychological and financial burdens, which impact caregivers' experiences of wellbeing, leaving them at risk of illness themselves (1).

Caregiver burdens are conceptualised and studied differently, which complicate the comparison of findings in studies and limit the use of recommendations for interventions across specific diseases. According to Bastawrous, it is important to distinguish between objective and subjective care burdens (10). Objective burdens are related to instrumental caregiving, as managing or assisting in use of technical devices, or practical caregiving tasks, whereas emotional burden and psychological distress are related to subjective experiences of the effects of objective burdens (10). Exposure to long lasting, demanding caregiver burdens may affect the wellbeing of caregivers through negative health effects and problems such as lack of healthy diet, sleep and exercise, which may put caregivers at risk (1).

Caregivers in chronic disease may also experience positive effects of caregiving. One reported benefit of caregiving included positive feelings engendered by being able to help another person, which tended to “give life meaning” (3). Adult children who provided care to a parent suffering from Parkinson's or Alzheimer's disease reported benefitting from caregiving based on its foundation in the parental relationship (11). Respondents reported positive feelings due to spending time with parents and having the opportunity to “give back” care.

Caregiving for dementia is associated with negative effects on caregiver health, and factors such as relationship to the patient, personal characteristics and gender may influence the impact on caregiver burden (12). Greater degrees of depression and stress and decreased subjective wellbeing in caregivers are associated with patient characteristics, such as impairments and the great need for care (3). In a study including patients with dementia and their caregivers, Prorok et al. found that the

caregiver burden in dementia related to lack of appropriate services, poor communication between health professionals and caregivers and lack of knowledge about the illness course. The authors concluded that the health care experiences of caregivers are less than optimal, and there is potential for improved care in several areas (13). In an Australian study, caregivers in neurodegenerative conditions, such as motor neurone diseases, multiple sclerosis, Parkinson's disease and HD, were asked what would prevent them from continuing to provide care at home, and the majority of respondents across diagnoses answered health issues such as depression (14).

The burden of caregiving for patients with severe cancer has similarities to caregiving in dementia. In a case study, Bevans and Sternberg reported that family caregivers tended to modify their lifestyles and give priority to the patients' needs, which often resulted in health related problems, such as sleep disturbances and fatigue (15). A review of interventions to support informal caregivers in the later stages of disease suggested that some interventions designed to support the caregiver directly, might decrease psychological distress and improve quality of life. Little is known of the effects of indirect support on the health issues of caregivers (16).

The caregiver role and characteristics of the caregiving course in different diseases may have similarities across chronic conditions. Caregivers are vulnerable to health related problems associated with the caregiver burden. With a goal of improving health care delivery and support family caregivers providing for care throughout illness course, knowledge of the caregiver's situation and specific needs is necessary. Therefore, the focus of this thesis is on family members who provide care for a family member with HD.

## Establishing partnerships

Roussos and Fawcett describe *collaborative partnerships* as a public health strategy for improving community based health services. Collaborative partnerships require contributions from multiple sectors, including health politicians, health professionals and patient organisations. An underlying assumption is that solutions to shared



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challenges rely on contributions from all actors involved. At a lower level, health professionals, patients and family caregivers may be collaborators in care-course partnerships (17). Levine et al. argue that family caregivers should be included in collaborative relationships as a work force, treated as important partners throughout the care process, and that family caregivers and their potential contribution should be given consideration (4).

Collaboration represents a core element of health care. Gaugler et al. use terms such as partnering and partnership in their discussion of conditions for collaboration between health professionals, patients, family caregivers and family members (5). They postulate that health care, based on partnerships between health professionals and family caregivers can be an effective strategy to involve family caregivers.

Partnerships based on collaboration should be based on mutual recognition and respect, open communication and addressing family caregivers' needs in the family context (5). Gaugler et al. further argue that family caregivers and other family members should be able to choose conditions for participation and own responsibilities in the care process. They suggest that a focus primarily on the family caregiver who is most responsible for providing care implies a danger to obscure a complex situation for other family members and their reasons for involvement or not in caregiving (5).

Family caregivers and health professionals may represent different values, beliefs and knowledge, which may lead to conflicts (18, 19). A recent study among family caregivers in severe mental health conditions revealed that family caregivers were reluctant to communicate with mental health professionals because they feared being excluded from collaboration and support due to conflicting dynamics between involved parties (20).

Providing care in HD is demanding because of the complexity of the disease, including physical and mental symptoms, characteristics as genetic nature, slow progression and enduring need for care throughout a life span. Family members and health professionals may need to collaborate over years. In order to promote collaboration and partnerships in health care delivery in HD, family caregivers' experiences from

encounters with health professionals may provide important insights to preconditions for successful collaboration.

## Health care in Norway

The Norwegian health care system is semi-decentralized, with the government being responsible for specialised care in hospitals. The municipalities are responsible for primary health care, including services from nurses and general practitioners (GPs). Citizens are listed with a local GP who plays an important role in collaboration if specialised hospital services are needed. Health professionals and social workers provide for health care in the municipalities in patients' homes or in local health institutions for nursing and day care (21). The system is tax-financed, and most services are free at the point of delivery, with out-of-pocket payments and co-payment for some services.

The Centre for Rare Disorders (in Norwegian: Senter for sjeldne diagnoser) is a multidisciplinary national competence centre organised as part of the specialised hospital services. The Norwegian centre offers nationwide information, counselling and educative services related to several rare disorders, including HD, for patients, caregivers and health professionals (22).

In 2009, The Ministry of Health and Care Services and the Norwegian Directorate of Health established a competency network for community health care for patients with HD and their families. A collaboration led by NKS Klover Institutions (in Norwegian: NKS Kløver institusjoner, Norske Kvinners Sanitetsforening) resulted in the establishment of the National Competency Network for Community Health Services in Huntington's Disease. The network functions to develop competence in resource centres and care services for patients and families with HD (in Norwegian: Fagnettverk Huntington) (23).

The Norwegian Ministry of Health and Care Services has initiated and developed governmental reports and documents with information about programs and guidelines of health policy and health care services. A white paper about the Norwegian

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Coordination Reform, implemented in 2012, claimed that “proper treatment - at the right place and right time” should be the new direction in health care (24). The paper pointed out three primary challenges in the Norwegian health services: 1) the need to establish better-coordinated services, 2) the need for increased focus on limiting and preventing diseases and 3) the changing range of illness in the population. The goal of the reform is to improve the quality and effectiveness of health care services by increasing resources to municipalities and improve the coordination of care, obliging municipalities to take the responsibility for improving the coordination of care. An individual care plan (IP) is a recommended tool to contribute to a coordinated care course and adjusted individual health services for patients with enduring and complex needs due to chronic disease or conditions. Next of kin as caregiver may be involved in developing an individual care plan. The program states that the delegation of responsibility for coordination of the care process should be clarified by organisational roles with a health professional with knowledge of the case and tasks in the process across health care system levels as coordinator (25, 26).

According to Norwegian law, a patient's next of kin is “the one the patient wants as his or her related person to be involved in health care plans or health service delivery” (25). Usually the next of kin is a spouse, parent, adult child, sibling or partner, but the definition of next of kin does not exclude other close persons in a role as caregiver, as a friend or neighbour. Family caregiver is a term in use describing a next of kin involved in different family care tasks (27). Governmental documents underline the importance of supporting the family caregiver through the illness course, which can be long and arduous. It is underlined that family caregiving is a significant resource in collaboration with health professionals and for the patient. The need for valuing family caregivers’ efforts is emphasized and the significance of their contribution in caregiving to maintain caregiving level of today is highlighted (27-30). A goal for modern family caregiver policy in Norway is to make the caregiver role visible, equal and flexible by offering counselling, advice, respite and openly valuing caregiver efforts (29).

National programs and guidelines are developed to support professionals in their collaboration with parents to find solutions for how to meet children's needs for adjusted information or support (31, 32). Children are not supposed to fill adult caregiving roles and may need protection not to suffer from lack of care or safety due to parental illness (26, 33). Figure 1 below illustrates how involved actors and relationships between them, are featured as partners in collaboration for health care delivery in Norway (27, 29).

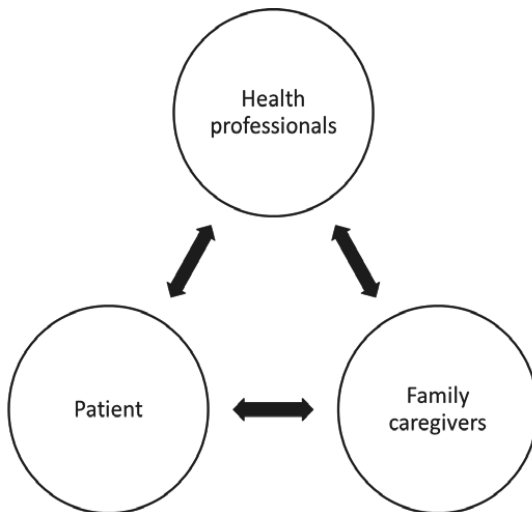


Figure 1

Health care systems do not have the resources to meet all care needs in the population nor family caregivers' need for respite. Health care systems will depend on contributions from next of kin and collaboration with volunteers to meet the increasing need for care. However, how health professionals may value and make the role of family caregiver equal and visible in collaboration is not clear. Is it possible for family caregivers to participate in caregiving on conditions adjusted to their needs and wishes in an everyday life? There is a possible tension in the interdependency in providing for care. Without clarity in preconditions for care and limitations in responsibilities, there might be a potential for exploitation of individuals as part of a work force.

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## Huntington's disease

In Norway in 1860, Dr. Johan C. Lund described patients who displayed involuntary movements similar to dancing. He named the condition “Setesdalsrykkja” after the valley where he observed the patients. Affected families were referred to as “chorea families”. HD was originally named Huntington's Chorea, after a physician named George Huntington (1850-1916) (34). In 1872, Huntington published an article in the journal *Medical and Surgical Reporter* of Philadelphia. He described a disease characterised by onset in middle life, which seemed transmitted from parent to child. He noticed a tendency for patients to develop mental changes, which sometimes led to suicide and movements (chorea) of the body similar to dancing (35).

### *Prevalence*

HD is a rare, hereditary and slowly progressive neurological disease. HD exists all over the world, but is most common in people of northern European origin (36). The prevalence of HD globally is estimated to 7-10/100,000 (36). A recent Norwegian population-based study suggests a prevalence of 5.9/100,000 in the South-Eastern Health Region in Norway (37). The Centre for Rare Disorders estimates that there are approximately 350-400 individuals affected by HD in Norway, with 1,000 persons at risk of gene affection and developing the disease (22). A juvenile form (onset under age 20) and a late form (onset over age 70) are well recognised. In a study from UK, Evans et al. found that the prevalence of HD is higher than previous studies suggested (38). This underestimation could have been the result of misdiagnoses or patients' failures to seek medical help due to shame, isolation or drug problems (39).

### *The genetic cause of HD*

HD is caused by a mutation in a gene called HTT (previously referred to as “IT-15”) at position 16.3 on the short arm of chromosome 4. In 1983, a link to the chromosome was first established, and the Huntington’s Disease Collaborative Research Group first identified the gene in 1993 (40). The abnormality of the gene is an expanded CAG trinucleotide repeat within a protein called Huntingtin (HTT). Striatum, a structure in

the brain, is involved in the regulation of movements and cognitive functions. A normal individual has approximately 20 CAG repeats, but if the number of repeats is above 36, there is a risk of developing HD. An association between the number of CAG repeats and age of onset has been found (41). Until recently, a myth persisted that the disease could not affect women (42). We now know that males and females are equally affected. HD is a single gene, autosomal-dominant inherited disease, which means that each child of an affected father or mother has a 50% chance of inheriting the gene and developing HD. In one family, all the children may inherit the gene and develop HD while in another family none of the children is affected (36).

It is possible to clarify if one carries the HD gene or not through presymptomatic genetic testing. However, genetic testing gives rise to ethical challenges related to sensitive information about family history, challenges in relation to other family members, questions of pregnancy and the fact that there is no cure for HD (43, 44). It may both be a relief and a burden to learn about ones carrier status.

### *Diagnosis and symptoms*

The diagnosis of HD is based on clinical symptoms and neurological findings and confirmed by genetic testing. In Norway, law regulates the procedures for the different forms of genetic tests. The results of genetic tests are highly sensitive and confidential with implications for siblings and children, as well as for the individual considering a gene test.

Age of onset varies, but two thirds of patients are diagnosed between the age of 30 to 55. The average course of the disease is 15–25 years. The slow destruction of nerve cells in the brain causes physical deterioration and a gradual progression of symptoms. Motor symptoms are characterised by involuntary movements (the chorea association) and impaired voluntary movements. Changes in brain processing related to memory skills and executive functions cause cognitive symptoms. Psychiatric symptoms include changes in personality and emotions. Depression is a symptom that appears to be part of the disease rather than a response to it (36). In everyday life, family members experience gradual deterioration in affected person's personality and skills,

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which influence the ability to maintain relationships and responsibilities and to participate in social life as before.

The progression of HD can be divided into five stages based on the Total Functional Capacity (TFC) scale, where the patient's scores are measured from 0–13. The scores reflect skills and abilities to engage in occupations, the capacity to handle financial affairs and domestic responsibilities, to perform activities of daily living and the level of care received (45, 46):

- Early stage (stage I): 11–13
- Early mid-stage (stage II): 7–10
- Late mid-stage (stage III): 3–6
- Early advanced stage (stage IV): 1–2
- Advanced stage (stage V): 0

HD affects persons differently within the same family, and patients will go through the stages at different times throughout the course of the disease, sometimes with overlapping symptoms and impairments (47). The progression of HD may also for simplicity, be divided into three stages:

*Early stage:* In the early stages of HD, people may notice subtle changes in mood, movement and cognition. The person with HD may still be able to drive and hold down a job but might require a little extra help doing these things.

*Mid-stage:* During this stage, people with HD will lose the ability to work and drive, and will need help performing activities of daily living. This is the stage where people tend to apply for disability pension. The movement disorder will create difficulties with balance, swallowing and voluntary motor tasks. Individuals will have increased difficulty organising and prioritising information. The behavioural symptoms will affect individuals differently, but irritability, aggression, depression and apathy at this stage can lead to personal and family issues, as well as the involvement of law enforcement.

*Late stage:* In the late stage of HD, patients require help with all activities of daily living. During this time, the person with HD may lose the ability to speak and respond but still be able to comprehend what is happening around them. Chorea may be severe or replaced by other movement symptoms, including rigidity, dystonia and bradykinesia. During this stage, many patients enter into long-term care facilities capable of providing 24-hour care.

### *Management*

There is presently no cure for HD, but medical treatment, palliative care and other services can support the patient's quality of life (40). The complex changes and needs in an affected person's everyday life and the great consequences of deteriorating functions necessitate a multidisciplinary approach including skilled health and social professionals with knowledge of the disease (47). There is ongoing international collaborative research to find ways to treat and slow down the course of HD and ultimately cure the disease. The European Huntington's Disease Network (EHDN) and the research collaborative, Cure Huntington's Disease Initiative (CHDI) Foundation, have ongoing research studies and working groups directed towards these aims (48).

As I have learned more about HD and its course, I have wondered about the great challenges and barriers family caregivers have to face living with this disease, incurable, rare and unknown for so many, yet with symptoms so severe, complex and care demanding.

## Caregivers in Huntington's disease

Family members may play a major role in caregiving for individuals with HD. The level of caregiving links to the severity of symptoms and functional impairments of the affected person. In addition, family caregivers in HD may be involved in multiple caregiving relationships due to the hereditary nature of the disease (49). For example, in one family, care may be required for a parent, sibling, spouse, partner, children and grandchildren over the years. Kessler featured the spouse as “the forgotten person” in families with HD (50). Research suggest that the characteristics of HD, such as its



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heritability, time of onset in family life and wide spectrum of symptoms, represent specific challenges for HD family caregivers that are distinct from caregivers of other severe chronic diseases (47, 51).

Care burdens cover physical and psychological burdens of care and experiences of wellbeing related to quality of health. Both qualitative and quantitative methods have been used to assess the impact of HD on family caregivers. The perception of caregiving as a burden suggests that HD may have a significant impact on the family caregiver's life and wellbeing. Aubeeluck and Buchanan have developed a quality of life questionnaire for caregivers (the HDQoL-C questionnaire), to measure life quality of spousal caregivers of HD patients. They assessed three domains of quality of life related to disease-specific factors: 1) practical aspects of caring, 2) satisfaction with life and 3) feelings about life (52). A focus group study identified four similar domains: levels of support, dissatisfaction with the caregiver role, practical aspects of caring, and feeling of emotional wellbeing. The study suggests that the quality of life is compromised in many ways for family caregivers within the measured domains and that own needs are negated as caregiving takes over (53).

Another study of quality of life among HD caregivers suggested that the number of hours spent on caregiving and lack of familiar support is associated with suboptimal quality of life (54). This resonates with Pickett et al.'s finding that family caregivers report less depression if they feel that they have control with their caregiving (51). In a study of predictors of quality of life, O'Connor and McCabe found that mood and satisfaction with social support predicted quality of life in family caregivers in HD (55). Semple explored the impact of HD on the family in a qualitative study and found that living with HD is stressful and emotionally damaging to all associated family members (56).

In a recent study of caregiver burden in HD, Banaszekiewicz et al. found that identifying those aspects of caregiving that influenced on family caregiver burden the most was difficult due to the great variations in symptom complexity and disease progression of HD (57). Nonetheless, some factors that influence family caregivers'

wellbeing were highlighted in qualitative and quantitative studies, including lack of knowledge of the disease, lack of social support and concerns for children at risk of the disease (57). Etchegary interviewed family caregivers and found that they encountered lack of knowledge among health professionals when they approached health services for advice or support, probably because HD is a rare disease (58). This finding echoes a study by Skirton et al. who found that lack of knowledge about HD is a problem among health professionals and the public (59).

Research also suggest that family caregivers in HD experience isolation from social activities and life outside the home (60) and that caregivers' needs for respite from caregiving are unmet (61). Williams et al. found that HD tended to dominate family caregivers' time. Some caregivers found it difficult to discuss the need for support with other family members. The family member's need for care often limited the caregiver's time for social contact with others and increased his or her sense of social isolation. They also found that family caregivers tended to give priority to caregiving over their own needs. Reasons for some families tending to isolate themselves from friends were concerns for children at risk of HD and the complexity of family relationships as difficult emotional disturbances (62, 63). The increasing focus on the need for multidisciplinary and interdependent long-term care tasks in HD has led to studies calling for improved, coordinated care involving family caregivers (58, 60, 64, 65). In a recent study, Wilson et al. suggest a model for care in Huntington's disease that consists of a triad of care comprising the patient, family caregiver and a health- or social professional in a role of key worker. They suggest that a key worker represent continuity and knowledge about the affected family's history of the illness course and need for health care services from a multidisciplinary team (66). However, there is a lack of studies exploring the family caregiver role in the coordination of care and interaction between health professionals and family caregivers in HD.

Personal concerns for family caregivers in HD seem to have similarities across national borders. In a study from the UK and the US, the authors suggest that experience of burden amount may differ related to culture differences and priorities of

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health service delivery, but still affect important personal issues as relationships, social support and emotional and practical life (62).

## **2. AIMS OF THE STUDY**

The overall aim of this study is to contribute with knowledge to promote collaboration and partnerships between health professionals and family caregivers living with HD.

The specific aims of this thesis were to investigate:

- family caregivers' experiences with the impact of HD on the family structure, dynamics and roles in the family
- coping strategies and behaviour patterns used by family caregivers to care for themselves while caring for a family member with HD
- family caregivers' experiences and expectations concerning collaboration with health professionals

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### 3. THEORETICAL PERSPECTIVES

In this chapter I present three theoretical perspectives I have found helpful for highlighting possible preconditions for establishing partnerships between family caregivers and health care professionals. These are theories dealing with family systems, coping strategies, and relational coordination.

#### Family Systems Theory

*Family Systems Theory (FST)* represents a shift in the perspective on the patient. From focusing on the individual, FST regards the patient as a part of a social family unit. FST was introduced in child health care and has gradually been used in different diagnoses, chronic conditions and in rehabilitation (67, 68). The origin of Family Systems Theory (FST) is *General System Theory (GST)*, introduced by Ludwig von Bertalanffy in the middle of the 20<sup>th</sup> century. His interest was the theoretical challenges and scientific problems related to understanding complex and interactive systems. Bertalanffy defines a *system* as a complex of interacting elements. Interaction means that the elements stand in a certain relation with interdependencies to each other (69, 70).

FST considers the family unit as one unique, dynamic and emotional unit that operates as a system of interrelated individuals in interdependent relationships. Families are adapting to changes using strategies that may be different from other families. The growth and development of each member depends on the others. Family members react differently to life events the family faces. Any change in the life of one family member affects behaviours and emotions of the other family members. There is also a notion of family progression through a family life cycle (71).

Within FST, the family unit is the primary social group for an individual. There are, however, many ways of defining *family* and the notion of family is ambiguous and influenced by culture (72). Some definitions highlight structures or relationships between family members, while other definitions focus on family patterns and

variations in cultures and changes over time. In our part of the world new types of family structure have appeared, structured by several kinds of new relationships among family members including non-biological relationships. Mixed marriages, one-parent families, lesbian or gay couples, fostering families and families comprising stepparent or -children are some examples. Differences in relationships and culture of a family make each family unique. Certain patterns and behaviour may have positive consequences in one family but opposite in another. Stereotyping families based on certain characteristics, as symptoms and behaviour patterns in an HD illness course may be a pitfall (72).

Within a framework of FST, Wright and Leahey have developed a concept for assessing family patterns and family life. *Calgary Family Assessment Model* (CFAM) is a theoretical concept consisting of three categories including 1) *structural* issues, with subcategories as gender, composition, social class and extended family, 2) *developmental* issues, with subcategories as stages of family life span and attachments, and 3) *functional* issues, with subcategories as roles, activities and problem solving. These categories contribute in different ways in establishing a healthy family life in a social context, or as limitations in the process (73). The authors define *family* based on how the members identify themselves; that is, beyond traditional and limited boundaries, such as gender, biological ties or legal citizenship (73, 74). I shall apply this definition in this thesis.

According to Morgaine, *family centred approach* means to take into account basic components of FST, such as how family members interact in patterns in relationships. A family unit's boundaries can be viewed on a continuum from open to closed, and internal rules shape family members (71, 75). Cohesion as an emotional bonding and adaptability are characteristics of a family system, and models for measuring family functioning have been developed (76). Metha et al. studied the applicability of FST to patients with severe cancer in palliative care (77). They suggested that a family centred approach is appropriate for clinical and research practice within palliative care. They also pointed at the need for accuracy in defining what "family" means, because studies sometimes refer to the family as a unit but include only one family member in the

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research. In the process of developing practice concepts for working with families, there are variations of terms in use, inspired by specific disciplines or purposes. Bell suggests distinguishing between family centred care and *family nursing*, declaring that family nursing is more than family centred care (78). She highlights preferred qualities of relationships in family nursing, interventions enacted through therapeutic conversations in a nurse-family relationship. Family centred care, she suggests, is more oriented to assessment of structures.

Participants in our study all belonged to a family unit and were related as family member to a person affected by HD. As a hereditary disease, HD has impact on family members and extended relatives through a family life course, sometimes involving several generations (47). I find the term “family centred approach” appropriate for the purpose of this thesis. I have chosen the concept of CFAM and the definition of family within a framework of Family Systems Theory relevant as perspectives to understand family patterns and dynamics related to HD affection.

## Coping and coping strategies

Coping concerns how individuals manage challenges and stressful events in their lives. Individuals perceive stressful events differently. Coping and stress can be defined in different ways depending on the theoretical perspective. Stressful events or *stressors* may be divided into chronic and acute stressors. Chronic stressors are “threatening events that have relatively long duration and no readily apparent limit” (79). Enduring caregiving demands in severe chronic disease, such as in HD, is one example of a threatening chronic stressor.

Lazarus and Folkman define *coping* as “constantly changing cognitive and behavioural efforts to manage specific external and/or internal demands that are appraised as taxing or exceeding the resources of the person”. External and internal demands refer to major happenings or everyday hassles that evolve in the relationship between an individual and the environment, perceived as stressful (80). In an individual perspective, internal resources include a person’s values and beliefs, which implicate

that gender, culture and age are factors with influence on choices for coping strategies (81). The process of coping can be described as a stepwise cognitive process of appraising the meaning of the stressful situation to a person (80). Researchers are increasingly giving the role of social support in coping processes attention (81, 82). Lazarus and Folkman's two main categories of coping strategies are *problem-focused* and *emotion-focused coping* (80).

*Problem-focused coping strategies* are efforts to solve or change the stressful event itself. As a first step in the process, a person will try to clarify the situation and identify challenges and problems. The goal is not necessarily to eliminate the problem; it may be a wish for control or limit of the problem. Clarifying a problem may, for example, reveal that the stressful event will be a permanent part of the person's life.

*Emotion-focused coping strategies* are techniques to regulate the emotional distress a condition inflicts by reformulating the impact or severity of impact in a person's life. Examples of emotion-focused coping strategies are to seek emotional support or try to reformulate the meaning of the stressor in one's life (80).

Coping concerns what individuals do, not the effect of coping strategies. Individuals may use both coping strategies from the categories to obtain or maintain psychological wellbeing in challenging situations. Nevertheless, the adaptive value of coping strategies varies. An important distinction is whether strategies tend to be helpful or unhelpful. In a given context, one kind of coping strategy may be helpful but might be unhelpful under other circumstances. Coping strategies may be complementary by enforcing each other and facilitate solutions for problems, whether it is to deal with emotional problems or practical challenges. The value of coping strategies is therefore dependent on conditions for coping (83).

The distinction of problem-focused and emotion-focused coping strategies is recognised as an important contribution from Lazarus and Folkman to modern research about coping (84). Carver and Connor-Smith suggest an additional third category of coping strategies, appropriate for understanding a broader aspect of individuals' coping efforts. *Dysfunctional coping strategies* are efforts based on denial



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of reality or ways of acting on a rewritten version of what is actually happening (85, 86). Use of dysfunctional coping strategies among family caregivers in dementia suggests that dysfunctional coping strategies are associated with anxiety and depression (87). How family caregivers cope with caregiving in HD will somehow influence his or her individual life. A strategy, which is helpful to a family caregiver's needs and preferences, may at the same time be experienced as worsening of conditions for the care recipient or family members in another family. Coping is a complex phenomenon and the individuals involved may have different perspective of helpful strategies.

One of the specific aims of my study was to investigate family caregivers' use of coping strategies and behaviour patterns. What are they doing to cope with HD while at the same time taking care of themselves? I shall apply the three categories of coping strategies of Carver (85), which seem relevant for broader understanding of use of coping strategies in a complex HD-illness course in a family.

## Relational coordination

*Coordination* is a concept central to collaboration between health professionals and family caregivers. Uijen et al. described the evolution of continuity of care and related concepts as the coordination of care (88). Up to the 1990s, collaboration and communication between different partners were essential elements in the definitions of coordination. Over the last 20 years, definitions of coordination have also included personal relationships (88). Coordination may unfold in various forms, and different concepts have put different weight on the role of structures, procedures and organisation of the collaborative practice for coordination of care (89).

HD develops in stages resulting in an increasing need for multidisciplinary care from medical specialists and health and social care professionals in the community. In addition to the HD affected person, family members affected by the disease may be in need for assistance from health care services, through several parts of their life span. Patient and family members enduring needs in a complex illness care course, changing

over time, call for a well organised collaboration with multidisciplinary health professionals in a coordinated care course (90). Collaboration may be organised through loose and casual connections between professionals and family members. Procedures for coordination may be structured through communication, for example by written programs. Suter and collaborators studied which competencies health professionals considered relevant for inter-professional collaboration. They highlighted communication as a main competency for collaborative practice and care coordination between providers of health services and patients and their families (91). The complexity of the needs and challenges in severe chronic diseases may necessitate care and support from multidisciplinary health professionals. Collaboration between health professionals from different levels of health care may involve challenging communication and delivery-of-care tasks. Fragmentation of health care may also occur, leading to requests for better coordination of care processes (89).

The concept *relational coordination* (RC) evolved from studies of coordination and safety of US airline companies' flight departure processes in the 1990s, and researchers began to understand coordination as the management of interdependencies between the people who performed necessary tasks (92). Relational coordination has later been used by researchers in studies of the health service (93), and may be useful for increasing the quality of coordination of care (94).

In a relationally coordinated care process, the individuals involved are regarded as competent partners, equally valued for their contribution in the process. According to Gittell, the concept includes three dimensions, which are essential for relationships between partners. These are 1) shared knowledge, 2) shared goals and 3) mutual respect (95). These dimensions reinforce four significant aspects of communication: a) Frequent communication helps to build relationships through the familiarity that grows between partners from repeated interaction; b) timely communication is crucial for quality in the delivery of care; c) accurate communication is important for updated information; and d) problem-solving-oriented communication is essential to joint problem solving. The combination of quality in the communication and dimensions of

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relationships between partners underlines the intention of RC to improve coordination of care tasks and enforce the relationships in coordinated work processes (94, 95).

The Norwegian Coordination Reform focus on the need for changes in administrative, structural and financial systems to meet demands for improvements in quality of health care services (24). Still, at the clinical level there may be challenges related to collaboration and coordination of care that may be understood within the framework of RC. RC adds specific relational qualities of communication and interaction between partners. The concept may contribute with new perspectives on family caregivers' experiences in encounter with health professionals.

This thesis is based on studies conducted in Norway, which has a health system with established structures for collaboration and organisation of health services. RC represents additional qualities of coordination of care and is relevant for the work in this thesis.



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## 4. DESIGN, SAMPLE AND METHOD

### Design

We sought to investigate the experiences and needs of HD caregivers from the caregivers' perspectives. We were interested in the experiences of family caregivers as individuals, living their lives in interactions with a person affected by HD. Caregiving takes place in the context of the home, foremost in a family system where family members are the individuals comprising the social life. Because qualitative methods are appropriate for accessing experiences of life events, a qualitative research design using individual, semi-structured interviews was selected to collect data (96).

### Sample

During the planning phase, we initially aimed to collect a purposive sample of participants with experience from caregiving to a person affected by HD (97). For ethical reasons we did not want to include participants who knew that they were carrying the HTT gene. With the assistance from medical departments in four different hospitals in Norway, we distributed 25 letters inviting adult participants older than 18 years with caregiver experience to participate in the study. We assumed that hospital units responsible for HD diagnostic services would have a name of contact persons or family members related to patients with HD; however, this assumption was too ambitious. We therefore extended our requests with the help of a community-based health authority responsible for nursing and follow-up programs for persons affected by chronic disease. The Norwegian Association for Huntington's disease and two specialised hospital departments were also helpful. We received 10 response letters via hospitals and specialised institutions, four through the lay organisation with the help of snowball method (97) and one through primary health care in a municipality. All respondents were included.

The final convenience sample consisted of 15 participants (3 men and 12 women). All participants related to one or several HD-affected family members. In the following,

the term *family caregiver* is thus used to refer to a person who provide for care in the family unit. The participants' average experience of caregiving was 11.6 years, ranging from 1-35 years. Their experiences included care of an HD-affected parent, spouse, sibling or adult child with experiences from caregiving in all five stages of HD. Three participants had provided care for several family members, including parents, siblings, spouses or adult children. All the participants had been in contact with specialist and primary care. Further details of the sample are presented in Table 1.

**Table 1. Characteristics of participants (N=15)**

<b>Age in years</b>	<b>N</b>
20–29	1
30–39	0
40–49	6
50–59	4
60+	4
<b>Gender</b>	
Female	12
Male	3
<b>Family caregiver's position</b>	
Spouse	10
Ex-spouse	1
Child of affected person	4
<b>Second caregiving course</b>	
Yes	3
No	12
<b>Family caregiver has children</b>	
Yes	12
No	3

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## Methods

### **Data collection**

I established contact with potential participants by phone and email. Empirical data were generated through semi-structured individual interviews with open-ended questions (96). We developed a thematic interview guide based on three topics: 1) living with HD in the family, 2) managing living with HD and 3) experiences from encounters with health systems. In the planning process, medical specialists in a hospital department from different disciplines with knowledge of HD contributed to the interview guide. In addition, I invited three family caregivers to discuss how to establish a trustful interview situation and assist establishing a familiar vocabulary. Several participants found it difficult to make an appointment fit into the caregiving schedule. I conducted all the interviews.

Participants' efforts to conduct the interviews impressed me, and I scheduled the interviews according to their wishes and convenience. Six interviews took place in participants' homes, four in an office at a local health institution, three in a hotel room, one in a hospital and one in a ferry waiting room. I followed the interview guide with flexibility to follow up emerging reflexions or questions with subsequent participants (96). Some of the participants said they had never talked about their situations with another person. Each interview lasted 60–90 minutes. We did not plan for patients to be present during the interview session; however, one HD-affected mother participated in agreement with her son. I conducted all the interviews from October 2011 to February 2012. After the interviews, I noted reflections for possible further use. In agreement with the participants, interviews were digitally recorded, and I transcribed the recordings afterwards.

### **Role of theoretical perspectives**

In the early planning process, we discussed different theoretical frameworks and concepts, which at that point seemed relevant and touched topics like collaboration,

nursing in chronic disease, user involvement, empowerment and coping. As the research work progressed, more specific theories and concepts inspired and supported discussions of research questions, analysis and findings. For example, we found that all participants were family members of the person who had HD. Even if concepts of family theory, coping and forms of collaboration have been present all the time, particular theories of family and family dynamics seemed increasingly appropriate to understand the role as family caregiver.

Gradually, I decided upon three specific theoretical perspectives for the interpretation of data, corresponding with the research questions; Family Systems Theory (71) with the Calgary Family Assessment Model (73), coping, focusing Carver's model of three categories of coping strategies (84, 85), and Relational Coordination (95).

The theoretical concepts were resources that helped us discovering nuances during the analysis process. Writing the thesis, after the articles had been written and submitted, the theoretical frameworks became more prominent to interpret the findings and explore possible preconditions for establishing partnerships and coordination of a care course. The theoretical perspectives are therefore more pronounced in the paragraph presenting my discussion of findings in the thesis than they are in the articles. I consider this as an indication of academic maturation at this stage rather than as omissions at previous stages.

## **Data analysis**

The transcribed interviews constitute the source of empirical data for all the three studies presented in the three papers (I, II and III). Data were analysed with systematic text condensation (STC), a strategy for thematic cross-case analysis of qualitative data (98). STC recommends a stepwise analysis of qualitative data. Analysis started after all the 15 interviews were conducted and transcribed. We considered the amount of interview material manageable, and there was time for reflection between each interview. We assessed the interview material and concluded that it was sufficient to elucidate the research questions.



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In addition to myself, the main supervisor (JCF) and the co supervisor (KM) were involved in all stages of analysis. We started with reading the transcribed interviews. In a meeting, we agreed to the topics of the study and made an overall plan for the data analysis. We followed a procedure that ensured that all three had time to comment and participate in negotiations and agreed to decisions.

Before following the four regular steps in STC, we conducted a step (here called step 0) to establish and differentiate topics for the three studies (papers I, II and III). Step 0 followed the same procedure as described for step 1 below. We read all the transcripts for an overview of data with potential topics related to the research questions. The three topics we decided upon were 1) the role of family caregiver, 2) managing HD and 3) experiences from encounters with health care services.

Each topic served as a starting point for the four-step analysis process in STC for each of the different studies (papers I, II and III). Guided by the procedures of STC, our analysis followed the four steps: (i) Read all the material for an overview of data and identify preliminary themes while bracketing preconceptions, (ii) review the transcripts to identify meaning units and organise them into code groups developed from the preliminary themes, (iii) condense the content of meaning units within each code group by means of subgroups presenting the most important aspects of each code group and (iv) develop generalized descriptions and concepts for specific themes for each study (98).

I present an example elaborating the details of the process in the Appendix.

*(1) Total impression—from chaos to themes:* We read the transcribed material to obtain an overview of all the aspects participants described related to the topics for each of the papers. Preliminary themes developed in the first step were recognized and negotiated. The major position for interpretation was determined in advance through formulated research questions. At this point of analysis, we were inspired by, but not steered by the supportive theoretical framework. We sorted out, combined and organised themes and designated some for further analysis in the research process. As part of analysis, we aimed to bracket our preconceptions and not let our professional

backgrounds, clinical experiences, interests or gender dominate the interpretation and understanding of the preliminary themes (98).

*(2) Identifying and sorting meaning units—from themes to code groups:* In the second step of the analysis, we re-read all the material to identify meaning units related to aspects of the themes for each of the studies. Meaning units are text fragments in the transcribed interview material, which may elucidate the research question (98). All the meaning units were roughly organised and sorted into code groups elaborated from the preliminary themes established in step 1.

*(3) Condensation—from code to meaning:* In this step, the contents of the code groups were processed by organising the meaning units into a couple of subgroups to elucidate major nuances within each code group. A condensate was developed based on meaning units from each subgroup. To keep in mind that the condensates should voice participants' stories, each condensate was written in first-person. A quote from one participant was chosen to illustrate the content from each subgroup.

*(4) Synthesising—from condensation to descriptions and concepts:* In this step, data were reconceptualised to develop analytic texts. We formulated descriptive stories of the investigated themes from the condensates and grounded the empirical data with an eye to the transcribed interviews to validate that the text still reflected the original context. We developed subheadings representing the categories elaborated from the code groups to express the different parts of the analytic text.

## Ethical considerations

The Regional Committee for Medical and Health Research Ethics (REC South-East B, ref. 2010/2072) approved the study prior to recruitment and data collection. All participants received an information letter about the purpose of the study and a short presentation of the researcher they would meet in the interview situation.

I had been working as a nurse in clinical practice with health care for patients affected by HD a couple of years ago, and there was a chance that I had met some of the

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participants. I informed the participants of this possibility and reminded of the right to withdraw from participation at any point in the process, without reasons or consequences of any kind. We aimed for information from family caregivers, not the HD-affected persons (patients). All participants received written and oral information about the study and signed an informed consent document.

Processing information related to needs of another individual is a delicate matter. It is necessary to have in mind that participants are telling their stories, experienced and perceived in a specific context. Participating in the research study may have an impact on relationships between family caregiver and care recipient. For openness about participation, I choose to ask for patients' signature as well. Not all the patients were capable of understanding the information document due to HD affection and decreased cognitive functions.

Being a person at risk of HD is a sensitive matter that can severely affect a person's life. We discussed this related to inclusion criteria. It is a possible burden of role conflicts or emotional challenges related to inheritance and family history of HD. We decided not to include persons at risk of HD. One of the participants did not know of personal risk status, but still wanted to participate. We accepted the person's choice. I had established a network of local health professionals with knowledge of HD in case participants wanted further contact with health professionals.



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## 5. MAIN FINDINGS

### Paper I

*Røthing M, Malterud K, Frich JC. Caregiver roles in families affected by Huntington's disease: a qualitative interview study. Scand J Caring Sci. 2014;28(4):700-5.*

The objective of this study was to explore family caregivers' experiences with the impact of HD regarding family structure, dynamics and roles in the family. Data were obtained from semi-structured individual interviews with 15 family caregivers in families with HD. Analysis was conducted with systematic text condensation (STC), a strategy for thematic cross-case analysis of qualitative data.

HD had a strong impact on family dynamics in our study. HD gradually impaired the affected family member with influence on family members' roles in the family unit, disturbing relationships between family members and by transitions of responsibilities. Participants from families affected by HD in the late family life cycle reported different experiences of impact on family life than did family caregivers with onset of HD in the early family life cycle. In both situations, family caregivers had concerns for children or grandchildren and other relatives at risk of disease. Participants experienced conflict of roles as the need for care increased. They reported that the quality in reciprocal relationships between family members changed during the course of the illness. The caregiver role gradually overshadowed other roles as individual family members. Spouses experienced a gradual loss of the partner and some experienced the situation as the gradual onset of caring for 'another child'. Role transitions resulted in new parental responsibilities and fewer shared social activities. Some experienced problems due to reduced income. Family caregivers also described changed relationships between siblings. Different reactions among siblings towards the disturbed family situation and the caregiver role sometimes resulted in loss of friendship and changes in previously supportive environments. Some participants experienced a role transition of becoming main family caregiver as a child or

adolescent, compensating for impairments of an affected parent by taking on adults responsibilities. While caring for the affected parent, these young caregivers reported anxiousness about their own or their siblings' risks of HD. As adults, some continued in the caregiver role for one or more siblings. Participants described the sense of duty to caregiving as an emotional challenge mixed with guilt for being a family member without HD.

We found that the family network, including extended relatives, was vulnerable to fragmentation. Family caregivers experienced a gradual sense of social isolation. In addition to the experience of losing the affected family member, family caregivers reported difficulties staying in contact with family members who had moved out. Participants described how contact with other relatives was complicated by emotional reactions to loss and family illness history.

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## Paper II

*Røthing M, Malterud K, Frich JC. Balancing needs as a family caregiver in Huntington's disease: a qualitative interview study. Health Soc Care Community. 2015;23(5):569-76.*

The objective of this study was to explore the behaviour patterns of family caregivers and the coping strategies they use to care for themselves while caring for a family member with HD. We obtained data from semi-structured individual interviews with 15 family caregivers from families with HD. Analysis was conducted with STC, a strategy for thematic cross-case analysis of qualitative data.

We found that family caregivers in different ways attempted to balance the affected person's need for care and the caregiver's own need for personal activities of interest. Participants described how they gradually gave priority to caregiving and put their own lives on hold. Family caregivers used various coping strategies in attempts to adjust to the illness course and increasing need for care. In early stages of the disease, emotion-focused and problem-focused coping strategies were common. Seeking information about the disease was important, and participants typically consulted health professionals, such as the local general practitioner (GP), or other family members to find answers to their questions. As part of accepting the diagnosis, family caregivers tried to regulate information, adjusting it to their needs for privacy or openness about HD in the family. Some wanted to protect other family members from full knowledge of the characteristics of HD, such as its heritability, whereas others wanted schoolteachers to know about their children's special needs, for example. Family caregivers protected themselves from emotional burdens by regulating involvement and social contact with others. They also attempted to protect themselves from future challenges of symptoms in the later stages of HD through meetings with severely affected persons and other family caregivers. In the early stages, participants also described using problem-focused strategies to manage the need for care. They continued their own activities and tended to maintain social contact. As the disease

progressed, the conditions gradually changed. In the mid- and later stages of HD, the caregiving responsibilities gradually shifted. Maintaining personal activities became more complicated and began to compete with providing care. Participants reported managing the increasing need for care by decreasing their own valued activities. Some participants described having less time to socialize with friends and participate in regular physical activities, which carried implications for their wellbeing. Caregivers reported that maintaining their occupation was a highly valued activity that they did not want to lose. Socializing with colleagues, financial independence and feelings of being valued as an individual were significant factors.

At this stage, several participants described arriving at a turning point where coping strategies used in the earlier stages were no longer effective. They were giving priority to care and had put activities of everyday life on hold. The increasing limitations on their own activities were intensifying social isolation and a sense of helplessness towards managing further challenges. Participants experienced that HD gradually overshadowed the possibility of having “their own life”.



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## Paper III

*Røthing M, Malterud K, Frich JC. Family caregivers' views on coordination of care in Huntington's disease: a qualitative study. Scand J Caring Sci. 2015. doi:10.1111/scs.12212 (E-pub ahead of print)*

The purpose of this study was to explore the family caregivers' experiences and expectations concerning collaboration with health professionals for persons with HD. We obtained data from semi-structured individual interviews with 15 family caregivers in families with HD. The analysis was conducted with STC, a strategy for thematic cross-case analysis of qualitative data.

We found that family caregivers made various efforts to collaborate with health professionals. In the early stages, sometimes before the HD diagnosis was set, caregivers tended to approach the GP to discuss the affected family member's symptoms and changes in behaviour. Participants expected invitation to consultations, but some reported that they had to make efforts to be present and having to invite themselves. Participants felt their information was a valuable addition to what was provided by the patient, and hoped to contribute to a broader picture of the challenges in the family situation and other family members' needs. In early stages of the illness course, caregivers felt it was important to establish relationships with health professionals for later contact if needed. Participants had positive experiences exchanging information in the early stages. They valued continuity in trusting relationships with health professionals where they felt they could benefit from the professionals' knowledge of HD during changes in the care course.

Participants perceived clarity of roles and responsibilities as a crucial aspect of collaboration, especially in later stages of the care course. Family caregivers wanted to know whom to ask questions and which professionals were responsible for each type of care throughout the care process. Family caregivers found the organisation and structure for collaboration in the local health system unclear. In the later stages,

participants were involved in meetings with multidisciplinary health professionals. Some family caregivers were members of organised formal groups established in the community with regular meetings for collaboration. Participants described that they initiated processes for decisions concerning care services and experienced that health professionals did not always recognise their contribution of knowledge. Caregivers felt that the low frequency of regular meetings limited their opportunities to discuss actual changes in the affected family member's functioning. Participants clearly expressed the need for changes in health care services.

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## 6. DISCUSSION

### Methodological considerations

Qualitative methods are appropriate to investigate human and social phenomena such as experiences and expectations in the natural setting of everyday life. Several factors may affect this type of research. In a qualitative study, personal opinions, experiences and preconceptions will affect choices made in the research process. Attention to the role and positioning of the researcher is therefore important. In this section, I discuss reflexivity, internal validity and external validity, including consistency of aims, research design and findings, and transferability to other contexts (99).

### Reflexivity

Reflexivity concerns how a researcher's position, motives and experiences influence the research process and results (100). Below, I consider different aspects of reflexivity as they pertain to my professional identity as female nurse, my experiences from clinical practice, choices of theoretical perspectives and my role as interviewer.

In my clinical nursing practice, I had experienced inadequate health care services and limitations in collaboration with HD-affected persons and their families. The patients' functional problems were given priority in a home setting, with obvious consequences for spouses and other family members. We did not have procedures for systematic follow-up of family caregivers or other family members. Caregiver's needs and HD impact in the caregivers' lives were not often given attention, although their needs were obvious. Limitations of organisational structures and traditional nursing responsibilities in the hospital made it difficult to accommodate new perspectives and working methods in collaboration with health professionals across health system levels. To some degree, as part of a family unit, I could imagine the complexity of care burden and possible implications in an everyday family life, as expectations for family life and relationships of family members. My interests circled around improving quality of care delivery and a curiosity about how family caregivers appraised their

caregiving situation. As I was interested in family caregivers' voices, it was natural to start applying for participants through hospital departments. These were my influences when I discussed, planned and decided the study design in collaboration with my supervisor. In retrospect, I consider establishing an advisor group of family caregivers early in the planning process helped me focus on caregivers' voice and identify their perspectives when different from mine.

Alternative theoretical perspectives could have added valuable knowledge to the narrow field of HD research. For example, a social science approach might have focused on structural aspects of health care delivery and prioritisation. However, supported by the body of research that suggested a need for more knowledge about HD, we choose to study HD from the family caregivers' perspective.

As theoretical frameworks, the Calgary Family Assessment Model (CFAM) and Relational coordination (RC) were new to me. Both frameworks have previously been recognised for their practical use in health services research (73, 95). These frameworks add different perspectives to processes recognisable from my clinical practice. I have chosen Lazarus and Folkman's definition of coping (80), and Carver's model of categorising coping strategies (85) as framework for investigating how family caregivers tried to manage living with HD. The categories were helpful to understand the function of participants' coping strategies, especially in the late stages, when the challenges increased.

While health professionals were willing to distribute the invitation letter, they recognized the focus in the study but admitted that their contact with family members was limited in the hospital. I assumed that patients' health records would contain information about next of kin or name of family members, but this turned out to be a false assumption in some cases. Our participants were all in contact with local or specialised health care services. We considered that recruiting participants outside the health services would have been difficult although this might have given us a broader sample. I suppose my experience and knowledge of HD were valued and trustful factors for family caregivers when they assessed invitation for participating.

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Kvale and Brinkmann's reflections and procedures for conducting interviews inspired my approach to the interviews (96). Kvale and Brinkmann use the metaphors of the "miner" and the "traveller" to describe different approaches to the interviewer's role and the process of interviewing. A miner searches for knowledge and an essential meaning of an object or phenomenon. A traveller searches for new ways of understanding the phenomenon of interest as it may appear in different social contexts (96). My role may be comparable to that of a traveller. To gain new insight and understanding in my encounters with family caregivers, I encouraged participants to share their stories of being caregivers in HD.

My previous experiences and knowledge from the field represented both strength and limitations. My knowledge of HD and its consequences expanded my capacity to understand and further elaborate care descriptions and challenges of symptoms described by participants. As a traveller, I was able to recognise some of the hills my participants had to climb. I think that my follow-up questions during the interviews encouraged participants to share their experiences. One of the male participants told that it was the first time he had felt comfortable with sharing his experiences from an enduring caregiving process. He said it felt right because I had knowledge about HD and illness course.

Kvale and Brinkman argue that interviewing is a skill that must be trained and suggest that knowledge of the interview topic is required to pose good second questions to the interviewee's answers (96). On the other hand, my habits and routines over the years developed in an established health system might have restricted my attention to new places of significance or meanings of knowledge. I expected to meet caregivers with specific concerns, and I did. I also met fathers, mothers, spouses, children and siblings of affected persons, who were engaged in enduring caregiving with loyalty to affected family member and who appreciated the occasion to share their stories. I sometimes felt challenged by the possible different expectations of participants and me for the interview situation. It caused me to reflect on possible pitfalls in the balance between themes defined in the interview guide opposed to themes upon which the participant wanted to focus, such as discussing consequences of leaving caregiving duties.

## **Internal validity**

Internal validity is essential for the quality of qualitative research (99). Depending on the aim of the study, text from conversations or observations of persons related to the researched phenomenon may be adequate empirical data, consistent with the study aim. It is critical to consider if the method chosen actually highlight and investigate the research questions. Different data collection methods may thus be useful in qualitative research. In this study, we planned to talk with family caregivers to gain information about their experiences. We considered semi-structured individual interviews appropriate in our study because they are suitable for gaining insight into experiences and participants associations to themes, generating follow-up questions (96). Focus group interviews is another data collection method (101), but we did not find such group interviews to be appropriate in our study. The caregiver-advisors underlined the individual differences in experiences and hope for future among potential participants as a challenge. An advantage of focus group interviews over individual interviews might have been the potential for communication between participants. Participants' shared backgrounds, characteristics and experiences could perhaps inspire them to share their stories and talk freely with other members of the group (97). However, we chose to give priority to the benefits of individual interviews and a possibility to follow up personal experiences and concerns of individual participants.

Again, we considered focus group interview as appropriate method for aim of the study in paper III. Information from experiences of collaboration with health professionals may be less sensitive. Still, being a rare disease, organising a group of participants from all over would be costly, alternatively from the same local health region, participants possibly would have known each other. We did not want to introduce others experiences from care burden in late stages to participants still in early stage of HD. In addition, the participants were not always sure which level of health system or what kind of health profession the person they met in meetings for counselling and collaboration represented. In the analysis, it may be a challenge concerning validity.

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In the process of developing the interview guide, we sought assistance from three family caregivers I knew from clinical work some years ago. I consider this group's contributions to the interview guide and interview situation as strength. They provided suggestions for wording that would be familiar to other caregivers rather than perceived by the caregivers as academic jargon. For example, instead of using the phrase "coping strategies", the caregivers suggested asking "what do you do to live with HD"? An even closer attention to the nuances of wording may have enriched the perspectives of family caregivers in the interviews. Demographic information was recorded in the beginning of the interview session, before talking about experiences with living in family affected by HD and caregiving, and experiences from encounters with health professionals. In the first interviews, the structure of the interview guide helped me cover all the topics. Learning through the process, the interviews developed into freer conversations with questions more naturally integrated. Participants told about their situations differently, and my need for control decreased. Reflections written immediately following the interviews as field notes were useful in this process. The interview sessions endured from 60-90 minutes. I had prepared for two meetings for interviews with each participant, but this did not seem necessary.

STC is a structured and well-defined pragmatic procedure for thematic cross-case analysis, experienced as possible to conduct in a reasonable way, recommended for novice researchers (98). The stepwise, detailed and systematic procedure made the process of analysing transparent allowing for involvement and reflections between the two supervisors and me. Allowing theoretical perspectives to support the analysis, STC was appropriate for our study. The method holds an ambition to investigate variety and breadth of experiences from a social phenomenon (as in our case, caregiving experiences in a sample of 15 family caregivers) and accommodated our discussions of the theoretical perspectives. Furthermore, thematic cross-case analysis is manageable in a sample of our size. I conducted and transcribed all the interviews myself which gave me good overview and knowledge about the content.

The contributions from my supervisors in the analysis have been of significant value. Both are medical doctors and represent different medical specialisations, professional

experiences and domains of research. Main supervisor (JCF) has specific knowledge from clinical practice with patient and family affected by HD, engaged in national and international cooperation and research concerning the disease conditions. This was important in developing the interview guide and recruiting a sample of participants that represented experiences from all stages of the disease. In the analysing process, his clinical experience and knowledge of HD enriched the possibility of catching nuances in participants' experiences different from my nursing perspective. His knowledge and research experience of collaboration in health systems was of value, especially in discussions related to collaboration and coordination. Co supervisor (KM) has long and wide experiences from clinical practice as a GP working in the community. She is experienced from qualitative studies about other marginal groups of patients. Her contributions were useful in discussions of characteristics of family caregivers and understanding the meaning of context. Both supervisors have played an important role to ensure that each step in the analysing process was worked through thoroughly and negotiating interpretations, before we took the next step. They have continuously contributed with relevant theoretical perspectives, updated literature and research studies.

### **External validity**

The external validity of a study concerns the applicability of its results beyond the study context. *Transferability* refers to how findings in qualitative studies may be of relevance for other groups, individuals or if findings are applicable by giving new insight in certain context (99). The term is appropriate in qualitative research, as the term generalizable is appropriate for use in quantitative research (97, 102). The sample in this study included variations in participants' experiences, data from family caregivers with different positions as family members (such as parents or children). The participants comprised caregiving experiences from all stages of the disease. In the recruitment process, we did not ask for family caregivers with experiences from specific stages of the disease. A more narrow recruitment regarding stage-specific experiences could have increased a nuanced richness in the data related to adequate



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diversity and breadth from the care course and specific needs related to stages of the disease.

The number of participants needed for a qualitative study must be balanced against the richness in the data and quality of the interviews (97). Our sample consisted of 12 women and 3 men. Women still conduct the majority of non-professional caregiving tasks, as reflected in our sample. Malterud called it a “rule of thumb” that the sample should be large and varied enough to elucidate the aim (98). It would have been strength to have a sample with more men. Still, we found that our sample represented a broad variety of experiences, and the male participants included had experiences from all stages of HD. All participants told about encounters with health professionals, which allowed the caregivers to provide information about the perception of collaboration from their perspectives in the interviews. As HD is an inherited disease with early onset, age and timeline in family life are significant factors for variances in experiences. Variation in age also provided sample diversity in the types of relationships represented between caregivers and affected persons.

We consider the findings transferable to family caregivers in Huntington's disease in cultures comparable to the western context, recognising that the family caregiver's expectations of the caregiver role, their level of responsibility for care and their expectations about having “their own life” may differ among cultures. Some findings may also be applicable to family caregivers in other severe chronic diseases affecting family life with losses and changes in relationships. Family caregivers in diseases such as Alzheimer and Parkinson's have been compared to family caregivers in HD and found to have similarities in burden experiences (51), and emotional distress (103). Yet, differences to consider are disease onset, hereditary nature and duration of the care course in the family life span. Uccelli et al. suggested that family caregivers in multiple sclerosis experience impacts on family life and wellbeing similar to those in HD (104). They also reported similar communication difficulties with health professionals.

All participants were relatives of care recipients. In a situation where a neighbour or friend provides for care, the caregiver experience might be different because the person would not be facing concerns regarding the genetic complexity of the disease. In a discussion of external validity, participants' perspectives may hence be different from next of kin without familiar biological bonds. Our sample also did not include family members who were not involved in caregiving responsibilities. The illumination of their experiences and reasons for the lack of involvement could be a theme for future research, although recruitment challenges are likely.

We did not include experiences or expectations regarding collaboration from other partners than the family caregiver, as health professionals or the patient. Information about preconditions for developing partnerships from a wider perspective is therefore unilateral and limited. There is a need for further research of expectations of partnerships with significant other contributors concerning relations in partnership and quality of care for the affected person and caregivers.

## **Ethical issues**

Considering the sensitivity of family illness situation and a possible need for further contact with health professionals, I gave information about the possibility for such contact to participants. After one year, one of the participants sought help for contact with specialised health professionals to discuss genetic questions. In my role as a health professional, facilitating contact with medical specialists has previously been part of my work, and I gave the request priority. The request may highlight the importance of planning for possible reactions to sensitive themes in an interview study and involved participants may need time for reflection before they decide to act. It may also highlight the importance of facilitated access to health professionals with knowledge of HD.

Considerations of the confidentiality and protection of participants' privacy are important in qualitative research (96). We reflected on confidentiality and anonymity continuously during the research process related to the sensitive information we

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possessed about families and individuals. Being a rare disease, family conditions and individuals are easier to recognise, especially in small regions. All quotations used in the manuscript are accurate, but in a few cases, I have changed or omitted gender of siblings and familiar bonds in relationships to maintain confidentiality.

An additional ethical consideration has been the challenge of providing balanced presentations of family caregivers' descriptions. In my reflections of the complexity of the experiences presented, which made impressions on me, it seemed easier to identify negative than positive experiences. It is possible that a sense from clinical practice of not providing for sufficient or adequate care made me pay particular attention to areas with need for changes. According to Kvale and Brinkmann, it may be a pitfall to leave the role of researcher and respond as therapist or friend (96). Discussions with supervisors in the analysing process were useful counteracting that. Participants' life- and caregiver experiences are subjective and represent their version of qualities of relationships with health professionals. Health professionals' and patients' perspectives are not part of this research study. As such, it is a limitation for a comprehensive exploration of interaction. I chose to prioritize the perspective of family caregivers.

## Discussion of findings

We found that HD had a strong impact on the family dynamics and how the role as family caregiver was shaped within each family system. Family caregivers experienced conflict of roles. Some participants had experience with caregiving as a child. Participants reported difficulties when balancing caregiving with valued activities in their own lives. Family caregivers expected recognition as competent partners in relationships with health professionals. Participants considered clarity of roles and responsibilities crucial for collaboration in a coordinated care course. Below, I discuss the impact of these findings, focusing on coping with the progression of HD, special challenges for young family caregivers preconditions for developing partnerships with family caregivers, and challenges for professional practice, with a special view to the significance of the context in which caregiving unfolds.

## **Coping with the progression of HD**

Participants experienced complex emotions of guilt and shame related to not being at risk while other family members developed the disease. In families affected by HD, the genetic and enduring nature of the illness and the lack of a cure represent additional challenges for caregivers. Research suggests that adult family caregivers in HD experience depressed mood and decreased quality of life more than caregivers in other severe neurological diseases do, such as in multiple sclerosis and Parkinson's disease (55). Our study contributes with knowledge about the complexity of coping as a family caregiver in HD and the importance of family context to understand coping strategies in different stages of HD.

Paper I revealed that the impact of HD on family patterns and dynamics shapes family caregiver's role. Transition of roles became visible when the role as family caregiver conflicted with other roles and needs. Participants tried to maintain a normal family life by balancing tasks and responsibilities of caregiving with activities valued in their individual- and family everyday life (papers I and II). Our analysis demonstrated that the need for care gradually overshadowed family members' needs and own activities, and they had trouble with taking care of themselves in later stages in the illness course (papers I and II). Over time, they lost a partner, friend and co-parent. In addition, they lost parts of their social life. In order to manage the challenges of a family situation with HD, family caregivers tried to adapt to problems as they occurred with problem-focused as well as with emotion-focused coping strategies (papers I and II). Later in the course, these strategies would often be replaced with more dysfunctional coping strategies (85).

Participants tried to solve practical tasks and responsibilities with new routines and priorities, compensating for tasks the affected family member usually would have done (paper I and II). In early stages, their problem-focused coping strategies seemed to help them to adapt to the situation. Confronted with gradually increasing caregiving needs, they set aside their own activities of preferences. The need for care developed in later stages of HD and the environmental context for coping gradually changed.

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Earlier strategies seemed no longer adequate. Problem-focused coping strategies, such as taking short periods of sick leave from work, did neither add sufficient time for care nor triggered increasing support. Participants used emotion-focused strategies to regulate distress. The emotional support from colleagues was important to them, and was a reason for trying to keep on working in spite of caregiving tasks at home.

In early stages of the care course, participants' use of coping strategies was adequate, but turned to be dysfunctional in later stages. They tried to continue using the same coping strategies to maintain a balance between roles aware of decreasing their own well-being. Participants described experiences of helplessness or kind of resignation in finding new more adjusted strategies. To deny or avoid knowledge of HD would be examples of dysfunctional coping strategies (85, 87). In later stages, participants were selective about whom to involve and how to inform about HD in the family. They delayed or held back information of HD to other family members to avoid conflicts and appraised timing and openness about the difficult question of heritability of the disease (papers I and II). They tried to protect themselves from emotional distress by avoiding information about HD progression. Participants' experiences of not worrying in early stage of the disease contrasted to their descriptions of feeling worried and exhausted in later stages. They were overwhelmed of conflict of roles and did not know how to solve problems differently. This point in the caregiving course for some coincided with loss of social support from family members or friends.

We found that family caregivers struggled with finding adequate coping strategies in later stages of the disease, similar to findings from a study among family caregivers in HD in Malta (105). A qualitative study of the impact of HD on family caregivers' quality of life indicates that their needs were compromised with the need for care (53). Helder and co-workers explored coping mechanisms and illness perception related to quality of life and found that spouses of affected persons dealt with challenges by accepting HD and tried to solve problems as they occurred (106). In another study, adult caregivers used avoidance as a strategy often as refusal to acknowledge the severity of the disease (107). In a study among family caregivers providing hospice care, Wittenberg-Lyles et al. found that receiving or asking for social support from

family members or friends in the care process was stressful. The family caregivers were afraid of feelings of guilt and loss of control (108).

According to Lazarus and Folkman, *social support* enhances adaptive outcomes and plays an important role for coping (80). Deficient communication between family members and health professionals (paper III) is probably one reason for the imbalance of care needs and possibilities for work, another reason is that family caregivers did not know of or found they had access to appropriate resources. Family caregivers had not addressed how to prepare for increasing caregiving responsibilities and loss of social support. An implication of this for health professionals is to balance possibilities for timely support and information to caregivers about available coping resources, especially during the later stages of disease when the social network may be disrupted and coping strategies seem dysfunctional.

Conflicts between family members' needs and preferences in a family unit may occur due to counteracting interests. Coping strategies that appear helpful for the family caregiver may function adversely for the care recipient or other family members. Loyalty to one's family, concerns for children or sense of family responsibilities are all parts of a family life, which may complicate individual preferences (72). Supporting family caregivers through the illness course should include sharing information and discussing resources for possible new solutions to problems (73), recognizing their limited options of adequate coping strategies.

Participants did not manage to solve increasing conflicts of roles and their experiences of decreased well-being. Our study adds knowledge though identifying "a turning point" in the care course when family caregivers' use of coping strategies in early stages of the HD may turn out as dysfunctional in later stages. Our study also underlines the significance of continuity of support from health professionals from early to later stages of HD illness course. Education programs, counselling, and support should incorporate planning for future resources, and how to better prepare family caregivers' use of adequate coping strategies in later stages. There is a need for more research focusing on the effect of interventions such as education programs and

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respite from practical burden, to support use of appropriate coping strategies in different stages of the illness process.

### **Special challenges for young family caregivers**

Although they are few, participants had experiences from caregiving as a child or teenager (paper I). Their contributions of specific caregiving experiences add important knowledge about HD affection in family life. In retrospect, the young family caregivers told that they entered into functions, which the affected parent previously had managed (paper I). The children felt lonely and were concerned about how long caregiving responsibilities would persist. As young family caregivers, they took care of younger siblings night and day in addition to daily meals and household tasks. Some described that they had been responsible in periods for medication to the affected parent. Their concerns for the family were far beyond a normal level in family life. They tried to keep their anxiousness as a secret. They did neither have the maturity to comprehend the severity of the situation nor the age to collaborate with health professionals.

Research about young caregivers' conditions in HD is limited. Focus has mainly been on spouses or adults in role of caregiver (50, 61, 62, 109). A study of coping strategies among teens from families with HD demonstrated that young teens underused helpful services, such as HD support groups (110). They applied dysfunctional coping strategies, such as hiding their emotional strains, probably motivated by desires to maintain family harmony. Another study presents descriptions from the caregiving situation of young family caregivers in HD as a gradual process wherein priorities are given to the need for care at the expense of the caregiver's own needs and lack of legitimacy for the needs of young family caregivers (111).

Previous research suggests that young carers who were most in need of support, lived in family situations with less possibilities of normal parental support (112). Our analysis suggests that children as family caregivers experienced lack of support from family members and extended relatives. Consequently, there is need for special

attention to the impact of family dynamics and young family members function. In a family systems perspective this is an example of changes in roles between family members as part of family dynamics and HD affection (paper I). A family centred approach is a possible way to identify dysfunctional caregiving functions in a family unit (73).

*Parentification* is a term used for a role reversal in which children act as parents and serve parenting functions for other family members. The concept has previously been used for other parental illnesses and disabilities (113). General practitioners (GPs) have the opportunity to support children in their situation and support parents through counselling, but their possibilities are limited by the 'invisible child' in parents' consultations and time constraints on collaboration with other professionals (114). Hooper suggests that parentification in childhood may have negative consequences and lead to an insecure attachment style in adulthood (115). A study of attachment in families with HD suggests that growing up with a parent affected by HD may affect adult attachment (116). For young family caregivers in HD, the burden may lead to increased vulnerability in later care processes as adult family caregiver (paper I and II).

Young family caregivers are at risk of losing a formative period of their life. Current health policy in Norway aims at to inform about children's rights as next of kin in the health service (25, 26, 31, 32). Our findings add knowledge to the unmet needs of children who take on caregiver roles and adult responsibilities. Further research is necessary to illuminate the role of health professionals in supporting children in families affected by HD.

Our findings confirm and expand upon our preconceptions that health professionals should meet family caregivers' needs as an individual in a family unit, with attention to family dynamics and needs of other family members. The health service needs to focus particularly on children in caregiver roles.



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## **Preconditions for developing partnerships**

In addition to the practical burdens of care, changes in family dynamics and emotional conflicts had impact on previously supportive and trustful relationships between family members (paper I). Anxiety or fear of future illness and loss of the affected family member(s) made communication between family members and extended relatives more complicated. Family units became vulnerable to fragmentation as HD progressed, and family members' engagement in caring tasks varied. It could be difficult for family caregivers to receive support from other family members and to establish trustful relationships with health professionals (paper II). HD onset in early family life with young children or adolescents represented complex challenges not associated with HD onset in later stages (paper I). Participants with experiences from caregiving through the later care courses described how the emotional complexity and sense of isolation increased (paper I, II and III). Family caregivers experienced that health professionals gave little attention to their experiences and competence of impact of HD on family members and family life (papers II and III).

Previous research shows that family members experience extensive practical burdens in addition to emotional burden from losses. The genetic nature of HD and onset of HD early in the family life cycle seem to be major factors in family disruption, including severe impact on family functioning and high levels of conflict (49, 61). Williams et al. found that family caregivers felt isolated, and that emotional problems led other family members to disengage from discussions about the effects of HD in the family or the family caregiver's need for support (62). Aubeeluck and Moskowitz suggest that family breakdown and emotional distress seem to be primary issues in the family caregiver's situation, referring to the genetic nature of the disease and the prolonged disease process (47). Their study illuminates the importance of specific HD knowledge of health professionals (47).

Family caregivers perceive a general lack of knowledge about HD among health professionals in primary care, and family members and main family caregivers have difficulties in obtaining access to support from health professionals (58). Support from

a local GP was important and helpful for establishing open communication between family members in early stage with possibilities for contact later. In governmental documents, values for health care services and collaboration between health professionals and family caregivers are underlined. There is probably a lack of clarity of how values are to permeate clinical practice, concerning responsibilities in caregiving and expectations from health services to family caregivers and conditions for family caregiving (27).

Nolan discusses lack of consensus on the nature and purpose of caregiver support (18), arguing that attributes of the caregiver role and strategies for collaboration differ depending on whether a caregiver is valued as a resource for the family, a co-worker or a co-client. He suggests that regarding the family caregiver as a *co-worker* is the most appropriate designation. Procedures where health professionals and family caregivers make appointments for caregiving or distribution of care tasks are, according to Nolan, not satisfactory as a primary basis for determining relationships and valuing contributions from involved partners. Weimand et al. emphasised nurses' dilemmas concerning support for the patient or relatives in mental health care (117). Nurses felt that their main responsibility in care was the patient's needs. In competition with needs of relatives, the nurses gave the patient priority (117).

Participants in our study reported that health professionals did not acknowledge their competence and needs in the caregiver role. When health professionals' collaboration with family caregivers is ambiguous, family caregivers are vulnerable and omitted from partnerships. McPherson et al. found that family caregivers had better experiences with health services when they perceived that their knowledge was valued by health professionals and when structures were presented to facilitate exchanging processes between health professionals and caregivers (19).

Gaugler and co-workers argue that *partnership-based health care* should be based on acceptance and recognition of all parties' competencies and contributions, emphasising the need for assessment of family caregiver's context and partnerships. A key factor for partnerships between health care providers and family caregivers is a shared

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understanding of the meaning of *collaboration*. Family caregivers are competent sources on context-specific care needs and their own needs (5). Participants in our study reported that they felt neglected by health professionals, who provided care without discussing the appropriateness with family caregivers or did not invite them to consultations with the patient. Without knowledge of the caregiver's background, context for caregiving and personal needs it is not easy to understand and deliver adjusted appropriate health care. Health professionals should value access to family caregivers' competence to improve understanding and planning for appropriate interventions in partnerships.

In our study, participants experienced that HD deteriorated family life, changed and disturbed family relationships (paper I) and interfered with family caregivers' abilities to balance caregiving and their own activities (paper II). A family centred approach and continuous assessment through dialogue with all family members may provide access to possible changes in the context of family caregiving and improve quality of care for family members and family unit (73). Our findings expand upon knowledge about family caregivers' needs for establishing relational partnerships with health professionals to; maintain positive relationships between family members, to maintain a stable family life, and to counteract gradual family disruption (paper I and II).

An implication of our findings for health professionals is the importance of understanding the specific context for collaborative caregiving in HD. Furthermore, our findings suggest that health care partnerships require health professionals to recognise the caregiver's competence and knowledge of HD and effects on family life. Establishing partnerships and exchange knowledge in communication with the family caregiver is an important strategy planning for health care and an enduring care process.

Another implication of these findings is that health professionals need to include family caregivers and family members in early phases of the disease. Future research could investigate long-term outcomes of establishing partnerships with family caregivers at earlier stages of the disease and investigate family caregivers'

experiences of more intensive involvement from health professionals skilled in HD, in local health care services throughout the illness course.

### **Challenges for professional practice**

We found that family caregivers wanted to be involved in collaboration and expected invitation to consultations with professionals knowledgeable of HD (paper III). Participants emphasised open and mutual dialogues but did not perceive that their contributions were sufficiently valued (papers II and III). Some participants reported positive experiences from previously established relationships with local GPs based on exchanged information, which for some allowed for continuity in contact in the later illness course (paper III). We found that participants experienced unclear roles and responsibilities in coordination of care and collaboration encounters with health professionals. Formal structures were not always established or adjusted to the family caregiver's needs and did not correspond with the developing need for changes in health care.

There is a lack of research on the family caregiver role in coordination of a care course in HD. Still, an increasing interest in multidisciplinary long-term care in HD has led to a focus on coordinated health care, involving family caregivers (58, 65, 90). Skirton and Glendinning identified two main reasons for unmet needs among patients with HD and their caregivers (60): inadequate coordination of collaboration between all professionals involved and the family, and lack of attention from health professionals to patients' and family caregivers' needs. Simpson and Rae argue for improved structures in coordination of care as a necessary part of guidelines and standards of care in HD (118). They suggest introducing a *clinical coordinator* for HD as a response to the longstanding problem in obtaining health services across boundaries of medical, psychiatric and social problems health service units for families with HD (118). This idea corresponds with participants' expectations for clear answers to questions and trustful contact with health professionals in the health care system (paper I and II). A coordinator may relieve family caregivers from the burdens of

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dealing with unclear administrative or structural challenges, and may facilitate mutual dialogues and caregiver involvement.

Studies from the Netherlands present positive experiences with a multidisciplinary approach, using an individual care plan developed with health professionals, patient and family caregiver together (64, 119). The linking pin in collaboration was a trained nurse, skilled in communication, in a role of case manager with an opportunity for frequent contact between all three parts. Specialised central HD-skilled health professionals supported a local HD team (64, 119). Future evaluation and research will bring further information about the usefulness of the approach related to quality of health for family caregivers and patient. In UK, a *key worker* approach is suggested as part of a model for collaboration of a triad of care (66). In the Norwegian health care system, the role of *coordinator* in an individual care plan (IP) is comparable to some of the initiatives we see in other countries. Since 2001, patients in Norway suffering from severe chronic disease and in need of multidisciplinary care have had a legal right to coordinated health care services through the development of an individual care plan (IP) for health care services. Health-and social care professionals in primary care are responsible for processing the care plan, and patients and family caregivers should be part of all stages in the process to improve quality of the care adapted to individual needs (25).

Some of the family caregivers in our study participated in formal and structured collaboration with health professionals. Being frustrated with difficulties in understanding allocated roles and responsibilities, they referred to lack of adequate communication between members of the care group and professionals' lack of knowledge of actual caregiving needs and situation (paper III). Veenhuizen and Tibben argue that an improved care course in HD for the patient and family caregiver in an outpatient department depends upon all involved persons participating in planning and follow-up programs in a coordinated care course (119). Their study describes the significance of individual care plans for health care delivery. Patients and family caregivers had continuous and accurate contact with health professional

responsible for coordination, with flexible and easy access to support from central specialized units with knowledge of HD.

These experiences can inform the discussion of how to improve health care processes concerning HD, and probably other progressive chronic diseases involving comprehensive needs of care, in Norway. While improving organisational structures, one also needs to facilitate relationships that allow caregivers and professionals to communicate and to develop a shared understanding.

Gittell's concept *relational coordination* (RC) emphasises the relational aspect as crucial for managing interdependencies between individuals who are engaged in care tasks (95). Within this framework, shared knowledge, shared goals and mutual respect are foundational dimensions of the relationships, strengthened by timely, frequent and problem solving oriented communication. Participants in our studies reported that the frequency of meetings with health professionals was unsatisfactory. Changes in affected family members' needs occurred in periods without agreements for appointments or meetings. Without frequent and timely communication, possibilities for sharing knowledge were scarce. Weinberg and co-workers explored coordination between informal caregivers, formal providers, and the impact upon caregiver's preparation to provide and manage care at home. They found positive effects of RC on caregivers' abilities to manage care, and they suggest greater attention to coordination with informal caregivers (120).

Our study suggests that improved coordination of care requires that health professionals explicitly acknowledge family caregivers for their competences and involve them as contributors in partnerships. Clarity of roles adjusted to each caregiver's resources for managing responsibilities is crucial. As HD is a rare disease, health professionals in small communities are less experienced. Partnerships with special experienced health professionals with knowledge are crucial and may be promoted if health professionals emphasise continuity of care and share knowledge with family caregivers throughout the care course. Further research is needed on how flexible relational communication patterns between health professionals and family

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caregivers, adjusted to stages in the illness course, may improve the coordination of care.

My thesis has highlighted relational communication as an important precondition for involving family caregivers and improving partnerships with health and social professionals. Aiming to improve health care services, health professionals are responsible for continuous efforts to implement new knowledge from research, and systematically evaluate existing use of procedures and guidelines. There is need for improved efforts to identify families affected by HD and establish contact with local primary health care, GPs and specialised medical units for better adjustment to need for early mutual dialogues and information in an HD illness course. Additional opportunities may build upon existing organisational structures, and improve partnerships in multidisciplinary health care services to support the patient, family members and family caregivers affected by HD. The geography of our country and a centralised organisation of specialist competence of health care in HD might be a limitation for easy and flexible use of competence and collaboration across health care system levels. Establishing HD networks for professionals, including collaboration with lay organisations is a promising way to enhance knowledge of HD and to increase access to information.





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## 7. CONCLUSIONS

This thesis suggests that:

- Huntington's disease (HD) has great impact on the family caregiver's life as an individual. The family caregiver's role is shaped by the impact of HD on the affected person in the family.
- The impact on the family unit may render it vulnerable to fragmentation.
- The impact of HD early in the family life span may result in more severe and unwanted family dynamic conditions.
- In some families, children and young adolescents experience being family caregiver with parental responsibilities.
- Family caregivers use problem-focused and emotion-focused coping strategies in early stages of the disease, though experienced as dysfunctional in later stages.
- Family caregivers need support to take care of themselves.
- Family caregivers expect to be involved in collaboration and share knowledge with health professionals through mutual dialogues.
- Family caregivers are in need of coordinated caregiving processes with clarity in responsibilities.
- Family caregivers want to establish partnerships with professionals with knowledge of HD, characterised by flexibility for contact, continuity, accurate communication, recognition of family caregivers' efforts and knowledge of how HD affects family members and family situation.



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## 8. IMPLICATIONS FOR PRACTICE

- Health professionals should adopt a family systems perspective in the management of HD to understand family structures, roles and context for care giving.
- Family caregivers should be supported by professionals as individual family members with own needs throughout the disease course.
- Health professionals should consider family situation and family context for caregiving in collaboration with family members.
- Health professionals should seek to protect children suffering from caregiver responsibilities by supporting adult family caregivers and family members in maintaining parental responsibilities and caring issues for young family members. When appropriate, contact with existing health- school or social care systems ought to be established.
- Health professionals should acknowledge the family caregiver's efforts to balance the needs for care and maintain their own activities. Support and information should be tailored to caregivers' needs as family member in different stages of HD.
- Health professionals should recognise the family caregiver's competence in established partnerships throughout the care course.
- Health professionals should actively probe for the family caregiver's preferences of involvement in caregiving and seek clarity of roles and responsibilities to improve coordination of care.

## 9. FUTURE RESEARCH

The findings suggest that future research about family caregivers in Huntington's disease could explore

- Effects of family assessment and support in the early stages with a focus on internal and external family support in a longitudinal perspective
- Effects of direct interventions and support to improve family caregiver's wellbeing
- Effects and experiences from regular respite for the family caregiver's health in the later stages of the caregiving course
- Development of information and education programs sensitive to the adjustments of family members and family caregivers individual needs at different stages of the disease
- Long-term outcomes and experiences from establishing partnerships with family caregivers and family members at an early stage of the disease
- Outcomes and experiences from increasing collaboration between health professionals skilled in HD from specialised and local health care professionals to facilitate services
- Family caregivers' experiences from adjusted support at the different stages of the illness course
- Effects of access to resources for appropriate coping strategies in different disease stages and the care process

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**APPENDIX**



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## Data analysis with systematic text condensation

Below, I present details from the stepwise process of data analysis conducted with systematic text condensation (98). Examples are drawn from paper III.

Two collaborators (main supervisor and co supervisor) and I read all the material independently. In the first two steps, the main supervisor and I worked together. The co-supervisor was more involved in the third step, continuously informed through all our discussion. We had ongoing discussion and negotiation about the content of the code groups and subgroups. From paper III, the concept of RC influenced the analysis in step 3 and became an underlying perspective for exploration of the caregivers' experiences. Theoretical perspective may add surplus value to empirical material, and we appraised preconditions for RC as such.

Topic from step 0: Experiences from encounters with health services

*1. Total impression—from chaos to theme.* We read all transcribed material to get an overview of participants' experiences with encounters with health services.

Preliminary themes discerned in the first step of the analysing were written down without systematisation or pre-decisions for theoretical perspective. Through discussion, we agreed upon a couple of preliminary themes, including family caregivers' experiences from encounters with health care services and collaboration with health professionals. We discussed possible preconceptions related to this theme, and I recognised that my expectations for family caregivers mostly dealt with difficulties in collaboration. I found unilateral negative expressions in a category for “fighting the system” in the theme of “meeting with health services”. Awareness of this through discussion with involved supervisors resulted in a more nuanced picture after having re-read the material.

2. *Identifying and sorting meaning units—from themes to code groups.* In the second step of analysis, all the material was re-read. By means of negotiation, we established six code groups from the preliminary themes in step 1: being seen and heard, organisation of health services, coordination, collaboration, a reciprocal need for knowledge and the family caregiver's initiative.

Meaning units presenting aspects of participants' experiences were identified and roughly placed into these code groups. Working even more systematically with meaning units, we realized that the code groups were not sufficiently accurate and another review was necessary. All meaning units were still included but finally reorganised into these three code groups:

- Sharing knowledge
- Being seen and heard
- Clarity in health care delivery and responsibilities

3. *Condensation—from code to meaning.* In this step, the contents of the code group were processed, first by organising each of the code groups into subgroups to clarify different aspects within the code groups. For example, meaning units revealing aspects of the code group 'Clarity in health care delivery and responsibilities' were organised into three subgroups: to share knowledge with health professionals, experiences of unclear roles, and responsibilities, and planning for a care task.

I developed a condensate based on the meaning units from each of the subgroups. To keep in mind that the condensate should tell and represent participants' stories, I wrote the condensate in a first-person-formulation. Below is an example of a condensate from the subgroup “being invited and involved”:

“I had to push on to gain entrance in consultations. I wanted to be able to give our children some answers to symptoms I had observed over periods, and what it is all about for us. I have seen the changes but I was not invited to meetings. I felt forgotten and overseen by health professionals. It is as if I just bother them.



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The worst thing for me is not to be seen; I feel they do not listen to what I have to say or need. I had a good experience once when a specialist apologized and was willing to meet me later. I have to be with my husband because he is not always telling health professionals the version of implications as I see it. I find it trustful when I meet health professionals I recognise from earlier consultations who remember what I have said and understand the disease history of my husband.”

A quote from a participant was finally chosen to illustrate the content of the condensate from the subgroup:

“I had to push on to be with my husband at the meeting in the hospital. I had to be prepared to give our children some answers, and I have seen so much of the symptoms. But I was not invited. They had forgotten to write it in the invitation letter. But I wanted to be there, I had to push myself into it, I just had to be there. That day when he wanted to take his own life and the follow up from health professionals was poor.” (E4)

*4. Synthesising—from condensation to descriptions and concepts.* In this step, data were reconceptualised from the condensates to develop analytic texts from each code group, descriptive stories of the phenomenon in focus, grounded in the empirical data, with an eye to the transcribed interviews to validate that the text still reflected the original context.

In the result section in paper III, *Sharing concerns with professionals* is an example of a selected subheading.



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## Papers



**Paper I**

**I**



# Caregiver roles in families affected by Huntington's disease: a qualitative interview study

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## Caregiver roles in families affected by Huntington's disease: a qualitative interview study

**Aim:** The objective of this study was to explore family caregivers' experiences with the impact of Huntington's disease (HD) on the family structure and roles in the family.

**Methodology:** We interviewed 15 family caregivers in families affected by HD, based on a semi-structured interview guide. The participants were recruited through hospital departments and a lay organisation for HD in Norway. Data from the interviews were analysed with systematic text condensation.

**Results:** Huntington's disease could have a substantial impact on the family system, the shape of roles among family members and the hierarchical order between spouses, partners, and parents and children. The relationship between spouses and partners changed during the course of the disease. A reciprocal relationship was

difficult to maintain, as the role as carer overshadowed other roles. Children of an affected parent could compensate for impairments by taking on adult responsibilities, and in some families, a child had the role as main caregiver. The increasing need for care could cause conflicts between the role as family member and family caregiver. The burden of care within the family could fragment and isolate the family.

**Conclusions:** Huntington's disease has a major impact on family systems. Caregiver roles are shaped by impairments in the affected family member and corresponding dynamic adoption and change in roles within the family. Making assessments of the family structure and roles, professionals may understand more about how to care for and support individuals in their role as family members and caregivers in different stages of the disease and family life cycle.

**Keywords:** caregiver role, family caregiver, family dynamics, Huntington's disease, chronic disease.

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## Introduction

Huntington's disease (HD) is an autosomal dominant neurodegenerative disease with a prevalence of 5–10 per 100,000 (1). A child of a gene carrier has a 50% risk of inheriting the HD gene. The clinical symptoms usually start when the carrier is 35–55 years, but there is significant variation in age of onset. The disease develops in five stages over decades, and symptoms appear as characteristic involuntary movements, behavioural changes and decrease in cognitive function. At present, there is no cure for HD, but much can be performed to alleviate

symptoms (1). HD causes increasing impairments and need for care. Patients will often be in a long-term nursing home in the latest stage of the disease, but relatives and family members have important roles as family caregivers in earlier stages.

Huntington's disease affects families in several ways. As the afflicted family member gradually lose functions, the conditions for family life changes with concerns for the future, and an allocation of daily tasks between family members. When caregivers learn that HD is a hereditary condition, the question of children being at risk will arise. The diagnosis may thus serve as an explanation for a spouse, parent or grandparent's strange behaviour, but also represents new challenges for the future.

During the different phases of HD, family caregivers in affected families experience practical care burdens, decreased social contact, financial problems and psychosocial challenges (2–4). The need for health services of

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700

family caregivers in families affected by HD seems to differ from the needs of caregivers to patients with other severe progressive diseases, such as multiple sclerosis and Parkinson's disease (5). The differences are related to lower quality of life, a complexity of genetic questions and lack of knowledge about HD in the society and in the health services (6, 7). The role as family caregiver unfolds in the family as complementary to role as family member. According to the role theory, a person might experience being overloaded by expectations of too many roles at the same time. An individual might experience role conflicts when roles are difficult to combine or fulfil (8, 9).

Family system theory can be relevant for understanding family aspects of HD. Wright & Leahey (10) have developed a theoretical model or concept for assessment, intervention and care in families with chronic diseases. The model is based on system theories, conceptualising the family unit as a system, in perpetual change. A family unit is composed by the persons who feel socially related to each other, not necessarily biologically. Family constellations may change over time, but the role of family caregiver still unfolds within the unit, and it may be explored using perspectives from family theory.

A family system consists of a small group of inter-related and interdependent elements and is characterised by hierarchies, subsystems and boundaries (10–12). Boundaries might be helpful for a family unit in its protection of values and community; at the same time, there is need of a certain permeability as a corrective and stimulation in the unit's communication with the environment. A traditional family unit may go through a life cycle that develops in stages with characteristic events such as childhood, adolescences, adults leaving home, time to retire and for some grandchildren. Family members continuously interact in relational bonds that represent subsystems, such as the relation between mother and child, the relation between siblings or the relation between spouses. To maintain balance in the process of family life, roles are adjusted when a family system face challenges (10), and one family member's illness may thus have an impact on the whole family system. Family life develops in phases and the dynamics in the family system shapes and regulates family members' expectations to their different role functions. A better understanding of how HD affects family systems and how family caregivers experience and perceive their role could be useful for adequate support of families affected by HD (13, 14).

The authors of this article are trained in nursing and medicine and have clinical and research experience from community health care, general practice and specialised medical hospital work. We are familiar with some of the specific challenges HD patients and other marginal patient groups are facing. We wanted to conduct a study to explore family caregivers' experiences with the impact

of HD on the family structure, dynamics and roles in the family.

## Methods

### Design and participants

We have conducted a qualitative interview study and recruited 15 individuals with experiences as a caregiver in a family affected by HD. Participants were recruited through hospital departments and a lay organisation for HD in Norway. Participants included three men and twelve women aged from 20–67 years. The sample consisted of caregivers who had experiences from caring for affected parents, spouses, siblings or children in all five stages of HD (Table 1).

### Interviews

Data were obtained from semi-structured interviews with participants. The interviews were conducted by the first author (MR) in October 2011–February 2012. Participants were interviewed once. Each interview lasted 60–90 minutes. The interviews took place where it was suitable for participants, such as in their homes, in offices, at hotels or other places. A thematic interview guide had been developed based on input from HD specialists at a hospital department, patients and family caregivers. A group of three family caregivers, all spouses of affected husbands, were consulted about factors of importance for the interview situation. The interviews centred on participants' experiences as family caregiver, perceived needs, coping strategies and experiences with the health service. Emerging themes and hypotheses were explored in interviews with subsequent participants. The material was digitally recorded and later transcribed verbatim by the first author.

### Analysis

We used systematic text condensation (STC), a method for cross-case thematic analysis of qualitative data (15).

**Table 1** Characteristics of participants (N = 15)

Age in years	Gender	Relationships of caregivers	Have children
20–29	1 Female	12 Spouse	8 Yes 12
30–39	0 Male	3 Spouse/parent	2 No 3
40–49	6	Ex-spouse	1
50–59	4	Daughter/sibling	1
60+	4	Son	1
		Son/sibling	1
		Daughter	1

Average years of caring experiences: 11.6 years.



In this project, the method involved the following: (i) reading all the material to obtain an overall impression and bracketing previous preconceptions; (ii) identifying meaning units, representing different aspects of participants' experiences from their role as family caregivers and coding for these; (iii) condensing the contents of each of the coded groups; and (iv) summarising the contents of each code group to generalise descriptions and concepts concerning family caregiving related to HD. All authors read the material. The complexity of the way the participants experienced their role as caregiver, and the influence of family dynamics was thoroughly discussed. All authors participated in the further elaboration of themes and interpretation of findings.

## Results

We found that HD could have a substantial impact on the family system, the shape of roles among family members and the hierarchical order between spouses, partners, and parents and children. The relationship between spouses and partners changed during the course of the disease. A reciprocal relationship was difficult to maintain, as the role as carer overshadowed other roles. Children of an affected parent could compensate for impairments by taking on adult responsibilities, and in some families, a child had the role as main caregiver. The increasing need for care could cause conflicts between the role as family member and family caregiver. The burden of care within the family could fragment and isolate the family. We elaborated these findings below.

### *Transitions of family members' roles and changes in relationships*

All participants of an affected spouse or partner experienced that their relationship gradually changed during the course of the disease and as the need of care increased. They said that it was difficult to maintain a balance in their reciprocal relationship, and this loss of partnership was difficult, as pointed out by a woman with an affected husband:

What hurts me most is that you lose the one you loved. A complete different person is sitting in the dining room today, another person than the one who was my husband years ago. That is sad for me. I am more like a caring person now, not his spouse and loved one as I was. (participant C2)

Partners and spouses experienced that they had lost their equal partner and that the disease had brought 'a new child' in the family. They told about conflicting responsibilities and new role functions concerning caring for their partner and raising their children, practical tasks and work. Sometimes, they had to protect the youngest children against uncontrolled behaviour from the affected

parent, such as unexpected aggression or rude verbal statements. They had to make efforts to ensure that the youngest children were not alone with the affected parent during the day. Some participants also described conflicts about the family economy, as their partner was no longer able to share the responsibility due to cognitive decline. For partners who worked part-time, it was not easy to increase workload because of the need of care at home. Some partners received payments by the welfare system as paid family caregiver, thus reducing the need for professional home-based care. While a role as paid caregiver could help maintain a balance in the family system, it could, however, also result in lower income and social isolation.

Participants who had taken care of siblings with HD described it as the end of a mutual friendship, replaced by a complex relation based on a sibling's need of care and functions in a role of caregiving. Important bonds between siblings gradually broke down as the symptoms progressed. One participant had cared for her affected mother for years, while a friendship between the two sisters gradually changed when her sister developed symptoms:

Yes, there is a lot of frustration because me and my sister were best friends, super friends, sharing everything, clothes, boyfriends, and excitements, and now she is not here, and in this process I have not been able to establish friendship of my own. (participant C1)

In addition to losing a good friend and a supportive companionship, the continuous care process prevented her from establishing a social life friendship outside the family.

### *Family caretakers must adjust to mixed feelings*

Some of the participants who had grown up in a family with one parent severely affected of HD described taking on a caregiver role early in their lives. Their childhood and adolescence were filled by adult responsibilities and tasks adults usually do. Compensating for their parent's impairment, they took care of younger siblings, followed them to school and day care, did the laundry and made food, as portrayed by a woman who cared for her affected mother:

I could not tell about it to my father. I isolated myself from others, and in dark times, after I had put smaller siblings to bed, their schoolbags were packed, food for the next day made ready and the house cleaned up, I had ten minutes for myself. (participant E1)

Typically, teenagers would try to conceal the difficult situation by not disclosing their challenges to anyone, while at the same time, they felt a lack of support from adults or other family members. They felt desolate in

dealing with difficult challenges, such as medication issues and a parent's suicidal behaviour. Family caregivers in our sample experienced mixed feelings in the caregiver role as sense of duty and guilt for not being affected. For some, it resulted in bad conscience and devotion. Some of the participants described that the role of caregiver and responsibilities could cause emotional conflicts among family members, as underlined by one of them:

Our son moved out early, but our daughter who stayed at home was like a chopping block for her HD affected mother. When something went wrong she was always the one to blame. She must have suffered a lot. (participant A2)

#### *The family network becomes vulnerable and fragmented*

In some families, the disease led to stronger boundaries between the family unit, other relatives and the social world. The family network was fragmented and the family became isolated. Changes in family structure and fragmentation made families more vulnerable with a lack of family members to rely on when support was needed. It seemed difficult to reconnect with relatives after longer periods without contact. A man's story illustrates the complexity:

I was 8 years old when my parents divorced. Dad was always angry and we did not know of HD. I lived with my mother and three elderly siblings a couple of years, and then they moved out. I remember when dad came visiting us, we were afraid and thought he was an alcoholic. [...] We did not have contact with other relatives, just a little with an aunt. My mother never told me about grandparents. I know nothing about them. We got a message when my dad died in a psychiatric institution. Two of my siblings later got married and had children. We did not have much contact. All my siblings died within 15 years of HD. I seldom see the kids and have no contact with the spouses. Now I live alone and help my mother when she needs it. (participant C3)

Families affected by HD early in the family life cycle seemed to undergo more complex changes, as the disease conflicted with the question of raising and other responsibilities. The impact on family structures seemed less severe and the participants seemed more relaxed about changes in roles and family relations in families with late onset of the disease.

## **Discussion**

### *Validity, transferability and reflexivity*

Participants in our study were recruited through hospital departments and a patients' organisation. Our sample

consisted of people who were willing to be interviewed about their experiences, and there may be experiences that we might have failed to illuminate. We have not elicited the experiences of relatives who did not take on a caregiver role. Our sample represents a variety of experiences with caregiving in different stages of the disease, making our results transferable to caregivers in various families affected by HD. Three researchers with different backgrounds as health professionals have been involved in interpreting the data, which we consider a methodological strength.

### *Age of onset and phases in family life*

Our study demonstrates how HD challenges the stability in the family system, by changing family positions, role functions and tightens some families' boundaries. The impact of the disease may be very different from family to family, because HD develops in phases over years, and symptoms and impairments may vary. The disease enters into family life cycle at different stages and will have different impact on the family system. Family caregivers experience particular difficulties when HD occurs in an early stage of family life and if the disease coincides with parental obligations for children and adolescents. In our study, this stage of family life seems to be a peak period for complex role expectations and responsibilities between family members. This is also a period of less social contact, while increased social support might be helpful. When the onset of HD occurs later in family life, the disease does not seem to have the same impact on the parental subsystem in the family, as in the case of an early onset. However, also in later stages of family life, we found that caregivers experienced complex feelings of anxiousness and anger for the future of their children and grandchildren at risk. This is probably one of the major specific challenges experienced by caregivers in families with HD. Caregivers in families with other severe and chronic neurological diseases also experience burdens and strains (16), but concerns for next generation's health, and the history of caregiving for affected parent is present only when the disease is hereditary.

Caregiving for persons with dementia can be very demanding and lasts over long term. Onset of dementia may appear early (17), but normally occurs in a late stage of family life with different consequences for the role of caregiver. It is difficult to compare caring experiences, but one important difference between the diagnosis of dementia and HD is the heredity. Adult children of parents with dementia may have different resources for support as caregiver because they are not at risk, and they do not need to be anxious for the future of their siblings. Several of our informants had experiences of caring for two or more family members. Some of them cared for a spouse and an adult child in the same period,

describing a sort of a 'who is next' situation. Dementia, like Alzheimer disease, is more common and well known than HD, so the threshold for adapted and organised health services and understanding is probably lower. This highlights the unique complexity of the caregiver role related to HD and the need of research of comprehensible interventions on individual level.

### *Children and teenagers as caregivers*

Huntington's disease affects parental functions, because the ability to fill the parental role is decreasing during the course of the disease (18, 19). We found that children and teenagers felt that their own needs were put aside to give priority to caregiving of an affected parent. A child in a parent's role is called 'parentification' (20–22) and is a way a family and a child may adapt to a challenging situation. Parentification may suppress a child's own needs and may be destructive if a child, for a longer period of time, has to manage tasks, emotional issues and responsibilities for which they are immature (22). The problem of parentification is recognised among families affected by different diagnoses and social conditions, as cancer (23, 24) and substance abuse care (20). Multiple sclerosis is a severe chronic disease with similarities to HD. A recent study from Iceland (25) describes findings among young caregivers of a parent affected by multiple sclerosis with onset in an early family life, similar to ours. Experiences of being overloaded and immature for adult's responsibilities, putting their own needs of care aside, add to the burden of suffering. For health professionals, this might be a message of the importance of assessing the possible impact on each family member. The same study (25) documents a positive development for young caregivers later in life. Increasing social activity and support combined with less care seemed to be helpful for family caregivers entering a new life situation. Some of our participants struggled with lack of family and social support when another family member was developing HD and needed care. Our participants, who were caregivers as a child or teenager, also described relational conflicts later in life. Emotional responsibilities in addition to practical tasks are highlighted as the most risky type of burden because there is less openness about it (26). Some of our participants described their situation as frustrating, due to the silence of their family HD history and lack of competence in the environment. A recent study concluded that growing up in a family with one parent affected of HD appears to affect a child's adult psychological attachment (27). If one parent is affected and the other parent leaves the family, the children will be even more vulnerable for extensive caregiver burdens and neglect of their own need of care. Health professionals aim to work patient centred, but do not always adopt a family perspective or use methods to uncover a family situation (4). Spouses and partners experience burdens of care (6, 16,

28), but healthcare professionals also need to be aware that children and teenagers may have the role as main caregiver in the family.

Our study suggests that the situation of children and teenagers in families affected by HD deserves increased attention and that a family system perspective may be helpful in the management of patients and families affected by HD. Health systems are supposed to support and educate family members who are caregivers, to contribute with appropriate care and to enable caregivers to stay healthy. More knowledge about appropriate interventions, such as counselling and practical support to family members and the family caregiver, is needed, to prevent family disruption in certain vulnerable phases.

### *What does this study add to previous knowledge?*

We are not the first to describe the heavy burdens of care that family caregivers in families affected by HD experience (2, 4, 13) and that children and teens experience burdens growing up in families with HD (19). Our study adds novel insights about the significance of the context and circumstances in which the caregiver role unfolds. In a family perspective, where experiences of family history and the context of family life become frameworks for understanding, our study highlights that the caregiver role might be experienced differently and have different consequences in families, even if practical burdens seems to have similarities. Therefore, supporting a family caregiver also should include the need of support as individual family member. Our study pinpoints the similarities of care burden and need for caregiver support in HD and other diseases and highlights particular challenges in HD: the complexity of genetics, the onset of the disease in relation to stages in family life and a general lack of knowledge about HD.

### **Conclusion**

Huntington's disease has a major impact on the family system. Caregiver roles are shaped by impairments in the affected family member and corresponding dynamic adoption and change in roles within the family system. Health professionals should make assessments of the family structure and roles, to understand and reveal possible role conflicts or changes in family life. As the disease and the family life cycle progress, there will be a need for flexible care and support adapted to the individuals in their role as family member and family caregiver.

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### Author contributions

Merete Røthing and Jan C. Frich designed the study. Merete Røthing conducted the interviews. Jan C. Frich and Kirsti Malterud have been involved as supervisors, and all three authors read the data material and were involved in the analysing process and discussions. All authors have read and approved this manuscript.

### Ethical approval

The study has been approved by the Regional Committee for Medical and Health Research Ethics (REC South-East B, ref. 2010/2072). All participants were given written and oral information about the study and signed an informed consent form.

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## Paper II

II



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

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### 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 adaption 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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## Paper III





# Family caregivers' views on coordination of care in Huntington's disease: a qualitative study

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**Background:** Collaboration between family caregivers and health professionals in specialised hospitals or community-based primary healthcare systems can be challenging. During the course of severe chronic disease, several health professionals might be involved at a given time, and the patient's illness may be unpredictable or not well understood by some of those involved in the treatment and care. **Aim:** The aim of this study was to explore the experiences and expectations of family caregivers for persons with Huntington's disease concerning collaboration with healthcare professionals.

**Methods:** To shed light on collaboration from the perspectives of family caregivers, we conducted an explorative, qualitative interview study with 15 adult participants experienced from caring for family members in all stages of Huntington's disease. Data were analysed with systematic text condensation, a cross-case method for thematic analysis of qualitative data.

**Results:** We found that family caregivers approached health services hoping to understand the illness course and to share their concerns and stories with skilled and trustworthy professionals. Family caregivers felt their involvement in consultations and access to ongoing exchanges of knowledge were important factors in improved health services. They also felt that the clarity of roles and responsibilities was crucial to collaboration.

**Conclusions:** Family caregivers should be acknowledged for their competences and should be involved as contributors in partnerships with healthcare professionals. Our study suggests that building respectful partnerships with family caregivers and facilitating the mutual sharing of knowledge may improve the coordination of care. It is important to establish clarity of roles adjusted to caregivers' individual resources for managing responsibilities in the care process.

**Keywords:** family caregivers, health services, coordination of care, collaboration, chronic disease.

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## Introduction

Huntington's disease (HD) is an incurable, genetic, neurodegenerative disease, with characteristic symptoms including cognitive impairment, involuntary movements and personality changes (1). The disease is autosomal dominant, with a 50% chance of transmission to offspring. The prevalence is 7–10 per 100 000, and the mean onset of symptoms is 30–50 years. Gradually, a patient's impairments can affect family members and keep them

from participating and functioning in everyday life (2, 3). Multidisciplinary care has been recommended for this problem (4). Research indicates that caregivers face challenges when communicating with health professionals and that knowledge about HD is often limited (5, 6). A patient gradually loses cognizance of his or her situation and the needs of others, which creates a huge challenge for health professionals and family members (1). Guidelines recommend a multidisciplinary approach in caregiving to patients with HD and that health professionals take active steps to involve family caregivers to improve the quality of health services to affected families (7).

Family members play important roles as caregivers in families affected by chronic illness, and the demand for family caregivers is expected to rise (8, 9). Collaboration between family caregivers and professionals is essential for

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the caregiver's support and may help them endure caregiving tasks; however, this collaboration may be challenging if the roles in the care process are unclear (10, 11). Research suggests that unrealistic expectations from health professionals may place an additional burden on caregivers (12) and that the communication between family caregivers and health professionals is not always optimal (13).

Coordination of the healthcare delivery and caregiving may take place through various mechanisms (14). The use of technology, the formatting of organisational structures and the characteristics of care tasks have been emphasised in the literature (15). However, recently, the technical requirements of the work and the quality of the communication in relationships between members of a patient's care team have been underscored (16). Relational coordination (RC) is a theoretical concept for the management of interdependencies between the people who perform the tasks (17). According to the theory, communication and relationships are crucial in relational interdependent work processes, as in caregiving, underlying more technical tasks (17). Three essential dimensions of relationships between involved partners are proposed as necessary preconditions for effective coordination: (i) shared knowledge, (ii) shared goals and (iii) mutual respect for one another's contributions. Preconditions for high-quality communication in relationships are frequency, timeliness, accuracy and a problem-solving orientation. In a relational coordinated care process, the qualities of communication and the dimensions of relationships are mutually reinforcing each other. All partners involved are believed to make a difference with their knowledge and dependencies.

In 2012, a coordination reform was launched in Norway to improve the coordination or integration of effective health care for people suffering from long-term conditions. One aim is to improve health services through better coordination of healthcare delivery, involving patients and their families (18, 19). The authors of this study have professional backgrounds in nursing and medicine and have experience in clinical practice in community health care, general practice, specialised medical hospital work and research in the field of communication, marginalisation and complex health conditions. These experiences motivated us to learn more about the coordination of care between family caregivers and professionals for patients with HD. We therefore conducted a study to explore the experiences and expectations of family caregivers for persons with HD concerning collaboration with healthcare professionals.

## Methods

### *Participants and data collection*

We wanted to conduct an explorative qualitative interview study (20). Participants were recruited with help

from specialised healthcare hospitals, community-based primary healthcare centres and a patients' organisation for HD in Norway. Elements of snowball effect resulted in contact with four participants recruited through membership of the patients' organisation. The sample consisted of 15 participants (12 women and three men) aged 20–67 years. Adults who cared for person(s) affected by HD without risk of the disease were requested. The participants represented experiences from all stages of HD and served as family caregivers for affected family member(s), such as spouses, siblings and children. Some of the participants had experiences from caregiving for several family members from two or more generations. The average duration of the caregiving experience was 11.6 years (Table 1). An interview guide was developed with input from health professionals experienced with HD and three experienced family caregivers. Semi-structured, 60- to 90-minutes individual interviews were conducted by the first author in the period from October 2011 to February 2012. All interviews were digitally recorded and transcribed by the first author.

### *Data analysis*

The transcribed manuscripts were analysed with systematic text condensation (STC), a cross-case method for thematic analysis of qualitative data (21). All three authors read the material obtained from the interviews and were involved in the analysis. The four steps in the analysis

**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 caregiver have children	
Yes	12 (80)
No	3 (20)
Contact with health services	
In community health care	15 (100)
In specialised hospital care	15 (100)

were followed: (i) all the material was read to obtain an overview of the data and got an impression of themes from family caregivers' encounters with health services, bracketing our preconceptions and identifying preliminary themes; (ii) meaning units were identified in the texts, representing aspects of participants' experiences related to our research question, followed by coding into code groups; (iii) to clarify different aspects within the code group, each code group was divided into 2–4 subgroups, from which condensates were developed and illustrative quotations were identified; and (iv) descriptions of participants' experiences based on the condensates were devised. The author group had ongoing, thorough discussions about the development of themes and the choices made regarding code groups, subgroups and the final categories of results throughout the process of analysis.

## Results

Family caregivers tended to approach the health services hoping to understand the illness course and to share their concerns and stories with skilled and trustworthy healthcare professionals. Family caregivers believed their involvement in consultations with ongoing exchange of knowledge with competent, local healthcare professionals, familiar with their specific family situation, was critical for improved healthcare services. Family caregivers expressed unclear understanding of their expected contributions to the care process. Clarity of roles and responsibilities, especially in later stages of the care process, was believed to be crucial for collaboration. We will elaborate further on these findings below. Quotations have been assigned each participant's identity marker.

### *Sharing concerns with professionals*

Family caregivers articulated a need for help to understand the illness, its course and consequences. Some family caregivers initially had very little knowledge of the illness, as it had not been discussed or was not part of the family history. Others had in-depth personal experiences with family members affected by HD. Some did not know what to expect, whereas others approached the health services with numerous specific worries about their futures and about the patients' health statuses. Some of them described how consultations with health professionals at an early stage of the illness trajectory had prepared them for the challenges ahead and made them foresee the impact the illness could have on their dual role as family member and caregiver. The genetic nature of the condition and that it could manifest in children and other relatives were common concerns.

In addition, family caregivers articulated a more general and basic need to establish trustful relationships with

health professionals through dialogue and counselling. They invested in relationships with the health professionals through sharing their stories, views and concerns, so that they would have someone to turn to if something happened or an urgent question emerged. Family caregivers' initial collaboration with health professionals was partly focused on understanding the present and the future and partly on building relationships for future help and support. A wife was informed about the disease in the late phase of family life and shared:

I have now an explanation to his behaviour and a name of the disease. I have spoken with the general practitioner, and if or when time comes, I will contact him again, and that will be fine. For our children it is too late, but we have concerns and hope for the future of our grandchildren.(B1)

Caregivers who were spouses, parents, children or siblings reported that they were not involved in the ways they wished. Being involved, such as being informed and invited to participate in consultations and meetings with health professionals, was crucial to these caregivers. The participants mentioned two reasons for this. First, the caregiver may have limited insight into the illness situation and need assistance in the forms of practical and emotional support. Participants claimed that they felt health professionals had been unintentionally misled by patients about the caregivers' and other family members' needs for support because the doctors were limited to the patients' accounts of the situation. Second, participants reported that they had their own needs as caregivers and wanted to contribute their understanding of symptoms, behavioural changes and challenges. One participant, a spouse of an affected husband and a mother of three, took several initiatives to be more involved and better heard. She was convinced that her presence in consultations could make a difference:

I had to push on to be with my husband at the meeting in the hospital. I had to be prepared to give our children some answers, and I had seen so much of the symptoms. But, I was not invited. They had forgotten to write it in the letter. But I wanted to be there, I had to push myself into it, I just had to be there. That day he wanted to take his own life and the follow-up from health professionals was poor.(E4)

### *Meeting competent and respectful professionals*

Caregivers described the need for competent health professionals who were knowledgeable and skilled in treating HD, but who also understood how the illness could affect the family. This expectation was not always met. Although travelling to specialised centres was an option and something they were willing to do, the caregivers also underscored the value of competent local

professionals. Family caregivers articulated a persistent need for consultations with health professionals who were able to take notice of changes and understand the family as a whole. They also pointed to the importance of continuity in the contact with regular consultations. Seeing new health professionals who did not know the family's specific story was considered a burden. In addition, long intervals between consultations were considered a problem for the family caregivers:

In early stages, we had frequent consultations with a medical specialist at the hospital. He knew our history and we did not have to repeat our story every time. Now we have meeting only once a year. We need more often contact with a health professional who understand our situation.(D1)

Family caregivers reported meeting health professionals in different settings and arenas and described how coordination and communication within the health services were not always optimal. Some participants described positive experiences from their contact with individual health professionals, reporting that they received useful information or were guided to other health and social services for specific requests. At the same time, participants described difficulties in identifying how issues and needs should be disclosed to other professionals who were involved in providing care. One of the participants liked this type of experiences to being left alone and to find solutions without the help of health professionals, though these professionals knew the situation and could have been involved. A male participant, an experienced caregiver for his wife and daughter, reported being listened to, but later realising that his experiences and views had not been taken into account:

Health professionals who are responsible for services to my daughter seem to understand that I am experienced and they hear what I say, but all the time I have asked for someone who could take the responsibility, as a link between the health system and her. Sometimes they just send her a report from a meeting about decisions made for 4 months ahead. She cannot deal with appointments or understand agreements. Suddenly a decision was made about dust wiping. But there are other more important things she need. It is as if they do not listen to what I mean.(A2)

#### *Clarity of roles and responsibilities*

Family caregivers reported that roles and responsibilities between involved health professionals and caregivers were sometimes unclear, which made it difficult to the caregivers to know what was expected of them. A wife and mother of two children were confused because she expected someone in the community health system to

contact her after her husband had left the hospital. The communication routines of the system seemed to conflict with her needs for care and support:

I was told at the hospital that we would be contacted for further follow-up by health professionals in the community. Later, they contacted me and I was satisfied to learn that they kept their words. But it turned out not to be so easy. The health professional who called us said he was not supposed to be our contact and that we should be taken care of by another. Then there was summer holiday and nothing happened. So we don't know if a health professional from community health system or the hospital will be in charge.(C3)

The caregivers saw themselves as members of care groups for the patients. They shared that they sometimes had to take the lead in these groups to enforce change. One participant described a positive experience from taking the initiative to ask health professionals from the hospital to head a meeting in the community to share information about HD and to inform the local team about special considerations in the caring process. The caregiver described feeling relieved of the responsibility to inform others about the disease. Living close to the affected person in his or her everyday life, participants described feeling responsible to initiate increased healthcare services, which they felt should be initiated by health professionals. A woman who had cared for her mother for many years and now cares for an affected sister perceived a disconnection between her world of practical daily care and the care discussed in more formal multidisciplinary community health-group meetings:

I guess I am the one who have to take initiative and do something when my sister's need for care is changing. As an example, if she needs anything else in her house, I have to take care about it. A community nurse is coming once a time every second week, but my sister needs more help, at least once a week, in addition to a nurse taking care of medication. We have established a group with a medical doctor and health professionals from primary care. We have a meeting twice a year, listening to each other about my sister's needs. This has been good for something; I have started application for disability.(C1)

## **Discussion**

### *Methodological considerations*

The individual interview is appropriate for collecting data to explore individual experiences from life events and social phenomena (20, 22). We considered conducting focus group interviews (23) but wanted the opportunity to follow-up more closely with participants who shared experiences based on certain themes. Although

collaboration is an issue, health professionals were not interviewed. A one-sided perspective in understanding collaboration between multiple actors is a limitation, but a specific focus is the perspective of family caregivers, and we decided to focus on their experiences.

Researchers' gender, professional experiences and cultural background shape data collection and the research process (20). The interviewer had a nursing background, experienced from work in local communities and hospitals; this experience may have influenced the levels of attention paid to the interview content. For example, in the past, the interviewer worked with implementation of structures for individual care planning programs for persons affected by chronic conditions. The close involvement of the other two authors throughout the process increased our abilities to capture diverse nuances of family caregivers' experiences. We were familiar with the challenges related to symptoms and the changes of functions in HD, as well as the health system in Norway. This knowledge may have supported our understanding of the contexts within which the family caregivers lived their experiences and developed their expectations.

Our sample comprised 12 women and three men. Including a greater number of male participants may have enriched the data material, as participation in work life and responsibilities in the family may differ with gender. Different positions in the family and the caregiver's gender may trigger different needs and solutions for health services, which could affect collaboration (24, 25). Male participants in our study shared caregiving experiences from all stages of HD, including care for affected family members from two generations and contact with health services. The interview material on caregiver experiences was rich and diverse, and we consider our findings generalisable for family caregivers in families with HD in health systems with developed primary healthcare services. Findings in this study may also be generalisable for caregivers in families affected by other chronic diseases or conditions with regard to the acknowledgement of the caregiver role in partnerships to improve the coordination of care.

### *Knowledge sharing*

Family caregivers reported seeking help from health professionals in order to understand disease progression and consequences. They also reported making efforts to share information they considered relevant for health professionals. Previous research has documented family caregivers' needs for information in families with HD (5, 26). HD is a rare disease, and the lack of knowledge and experience among health professionals may present challenges (6). Research suggests that the difficulties in gaining access to information, poor communication and lack of interaction between family caregivers and health professionals are also experienced by family caregivers in

better-known conditions, such as Parkinson's disease, other forms of dementia and in end-of-life care (27–29). Compared to caregivers in families with other chronic conditions, caregivers in HD may be in a more vulnerable and complex situation. The characteristics of HD may have significant impacts on multiple family members over time in early family life. The rarity of the disease, its symptom characteristics, time for onset and genetic component necessitate knowledge sharing in the early stages of the disease, which could play a significant role in future collaboration and management of symptoms.

Participants reported having desires to share their knowledge, but feeling uninvited to do so by health professionals. In another study, caregivers found that health professionals did not exchange knowledge with informal carers, citing reasons such as privacy and confidentiality (13). Knowledge sharing may promote a common understanding of the situation and the challenges at hand and may reveal knowledge that family caregivers do not want or need to share with health professionals. While most health professionals may have general knowledge about HD, the contexts for the illness course and family histories differ and may need individual care and support (30). Mutual dialogues may promote knowledge sharing in the form of RC, which can serve as a framework where new understanding of changes and challenges based on shared knowledge can become a precondition for shared goals and for revising functional goals in the care process (17). Participants underlined a need for continuity in relationships with health professionals. Sharing knowledge over time with continuity in relationships might prevent the power imbalances or tensions that can create barriers between caregivers and health professionals (13).

Multidisciplinary care services tailored to the needs of the HD-affected person require coordination of interdisciplinary collaboration (4, 31). In addition, healthcare professionals must consider the possible differences in the needs of the caregiver and the needs of the patient (32). Professionals from multiple healthcare-related disciplines as physiotherapists, occupational therapists or pedagogues may play important roles in the care team in addition to nurses and doctors. Our findings indicate that health professionals should encourage the family caregiver to participate in consultations and should then integrate the caregiver's knowledge as part of a common understanding. Routines and procedures for including family caregivers may present opportunities for flexible ongoing contact adjusted to the disease trajectory and care process. Continuity in relationships should be given priority in the coordination of the care course.

### *Fostering mutual respect in collaboration*

Family caregivers expected respect from health professionals as competent partners in patient care. Our findings

also suggested that lack of communication and unclear expectations represented additional burdens for the caregiver. Our findings are in line with previous research suggesting that poor communication between caregivers and health professionals may lead to inappropriate care and place extra burdens on family caregivers (6). Lack of continuity in communication and coordination between partners was found to have negative impacts on patients and caregivers in another severe but more common neurological condition, Parkinson's disease (27). Research suggests that conflicts may arise between actors involved in care processes related to how they define each other's value of positions and knowledge. Family caregivers may challenge nurses as professionals because they are sceptical about releasing control (10). In a study of collaborative practice among health professionals, role understanding and communication were highlighted as two main competencies, while competencies such as a positive attitude and mutual trust were described as characteristics of individuals and not as competencies of collaboration (33). Ongoing mutual dialogues may increase understanding of role strains and the significance of shared knowledge (13). Mutual respect is aside shared knowledge an essential dimension of relationships in relational coordination. It involves an acceptance of the different but equivalent competencies and skills of the actors, which complement one another in collaboration. Mutual respect fosters receptivity to communication and contributes to the development of shared knowledge (34).

In our study, participants also reported poor communication related to infrequent meetings. Established routines for integration of caregivers' knowledge during the care course were not experienced as standard practice or as a guarantee for participants' experiences of being involved and acknowledged. Weinberg and co-workers applied the concept of RC in a study to assess coordination between health professionals and informal caregivers (35). Interaction along dimensions of quality and frequency of communication, as well as the supportiveness of relationships, was measured. The results suggested that relational coordination had a positive effect on caregivers' management of care and understanding of their roles. The frequency of meetings and accurate communication may not be standardised but must be adjusted to the illness course and family members' needs as caregivers.

RC may be helpful to improve coordination of care in HD, but the concept is in a relatively new stage of development, and further research on the strength of coordination in chronic conditions is needed (16). Further

research on how family caregivers may be involved in a coordinated care process with respect to their competences is also needed.

## Conclusions

Our study suggests that family caregivers should be acknowledged for their competences and should be involved as contributors in partnerships with health professionals to improve the coordination of care. Involving family members and family caregivers from early stages of the disease may give health professionals more appropriate information and knowledge of the illness situation. The clarity of roles adjusted to each caregiver's resources for managing responsibilities is crucial. Unclear roles in collaboration might be experienced as an additional burden and debilitate coordination of the care process. Health professionals should bring competent knowledge of HD-specific characteristics to encounters with family caregivers and should emphasise continuity in contact for sharing knowledge throughout the care course.

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## Author contributions

MR and JCF contributed to study design. MR collected the data. MR, KM and JCF were involved in data analysis and manuscript writing.

## Ethical Approval

All participants received an information letter and were given oral information about the study before interviews took place. They all gave written consent for participation and were informed that their opinions or statements had no implications for the care process or healthcare delivery related to affected person or themselves. The study has been approved by the Regional Committee for Medical and Health Research Ethics (REC South-East B, ref. 2010/2072).

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