Palliative care at home. Supportive needs of informal caregivers of patients with amyotrophic lateral sclerosis

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Dean

Memento

The fear of my own death is not so strong,
It's just the deaths of those who I adore.
How shall I live when they are no more?

Alone through fog I fumble deathalong
Get pushed into the dark while I'm obeying.
The leaving hurts not half as much as staying.

He knows it well who can identify;
– And those enduring it may please forgive.
Just think: one's own death one just has to die,
But with the deaths of others one must live.

Memento

Vor meinem eigenen Tod ist mir nicht bang,
Nur vor dem Tode derer, die mir nah sind.
Wie soll ich leben, wenn sie nicht mehr da sind?

Allein im Nebel tast ich todentlang
Und lass mich willig in das Dunkel treiben.
Das Gehen schmerzt nicht halb so wie das Bleiben.

Der weiß es wohl, dem gleiches widerfuhr;
– Und die es trugen, mögen mir vergeben.
Bedenkt: den eignen Tod, den stirbt man nur,
Doch mit dem Tod der anderen muß man leben.

Mascha Kaléko, 1956[1]

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[1] Kaléko, M. (1956). *Sämtliche Werke und Briefe*. München: DTV. English translation: Nolte, A. (2017) *Mascha. The poems of Mascha Kaléko*. Burlington/VT: Fomite Press. Reprinted with permission.

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This project would not have been possible without all the informal caregivers and people with ALS participating in the interviews throughout Switzerland, as well as the healthcare professionals. It is my sincere hope that their voices are heard through this research.

Summary (English)

Amyotrophic lateral sclerosis (ALS) is a fatal, neurodegenerative disease primarily of the motor neurons. ALS is divided into different subtypes which have different symptoms depending on the symptoms. As the disease progresses, people with ALS become more and more immobile, in need of care, and are less able to do everyday activities themselves. Around 600 people live with ALS in Switzerland. Informal caregivers, primarily family members, but also friends and relatives, the so-called informal caregivers, play a central role in accompanying, looking after and caring for people with ALS. Taking on these activities can be stressful for informal caregivers, so that different types of support may be required. There is only sparse information about the need for support for these informal caregivers in Switzerland.

The first chapter introduces the topics of amyotrophic lateral sclerosis, palliative and multidisciplinary care, and informal caregiving for people with amyotrophic lateral sclerosis. Subsequently, the second chapter lays out the research questions and objectives, methods, and methodology used in this thesis. The methodology of Critical Applied Ethics in empirical bioethics to connect the different strands of this cumulative thesis is also presented in this chapter.

Chapters three to ten form the results section of this cumulative thesis, whereby I first present an overview of needs of informal caregivers of people with ALS during the disease course in the form of a systematic review using narrative synthesis. This review synthesized needs relevant after diagnosis, across the caregiving course, and at the end-of-life. In addition to this corpus of evidence, the fourth chapter provides an overview of supportive needs of informal caregivers in Switzerland using a thematic analysis of interview data from both informal and professional caregivers. The chapter outlines several unmet needs of informal caregivers such as administrative burden, advance care planning early in the context of ALS, and underlines the importance of social support, be it in peer-groups or community care.

Complementing this largely descriptive research, the fifth chapter provides a grounded theory-based approach to understand informal caregiving during the disease and caregiving progression of ALS. Informal caregiving is understood as an upholding a steady state among different activities which informal caregivers perform. This process

of holding of the balance is the organizing element to four secondary categories which could disrupt or facilitate a balance. Organizing support could both shift the balance in favour or against the caregiver depending on the availability of support, being present could restrict or enable informal caregivers' activities. The categories of managing everyday life and keeping up with ALS further impacted the steady state.

The systematic review found a lacuna of research on bereavement needs of informal caregivers. Hence, the sixth chapter provides a thematic analysis of the experiences of bereavement of informal caregivers. The chapter discusses support after bereavement for informal caregivers of persons with ALS in the established public health model of bereavement support. Bereavement support needs to be proactive from healthcare professionals; however, it requires goodness of fit to address those in need and not those who are adequately supported by informal sources of support.

Before the death of people with ALS, they have to make decisions regarding their end-of-life circumstances. The seventh chapter discusses end-of-life decisions via brain-computer interfaces for people with ALS. Ethical challenges especially regarding the legal requirement to maintain physical control over the pharmakon in assisted suicide are presented in the context of ALS. The chapter concludes that physical control should not impede autonomous decision-making in ALS if people with ALS can communicate and have control via brain-computer interfaces.

The eight chapter outlines informal caregiving at the end-of-life in home care in a general manner. Informal caregiving at the end-of-life is blurred on several accounts: between being an informal caregiver and a non-caregiving close person, between formal/professional and informal caregivers, and between caring for others and self-care. This chapter is not specific for ALS but introduces the intricacies of informal caregiving through the lens of Care Ethics.

The ninth chapter discusses the conceptualisation of hopelessness of people from ALS. It is often assumed that hopelessness and demoralization impair decision-making on part of people with ALS. The chapter argues that in the context of ALS however, hopelessness is often conceptualized incorrectly. This essay won the 2019 Paul Wainwright Postgraduate Essay Prize.

Informal caregivers need different assistance and assistive technology from the state to provide their care. The tenth chapter identifies unfair conditions in the provision of assistive technologies and devices in ALS that arise because of a gap in the transition from disability insurance to old-age and survivors' insurance in Switzerland.

I conclude my thesis with a critical discussion and contextualization of the present research. Using Critical Applied Ethics, a thread is drawn that links the normative and empirical dimensions of the thesis. Subsequently, clinical and policy recommendations are proposed to improve the situation of informal caregivers of people with ALS.

Summary (German)

Die amyotrophe Lateralsklerose (ALS) ist eine fatale, neurodegenerative Erkrankung primär der Motorneuronen. ALS ist in verschiedene Subtypen unterteilt die symptomatisch verschiedenen Verläufe haben. Mit Progredienz der Erkrankung werden Menschen mit ALS zunehmend immobiler, pflegebedürftiger und können weniger Aktivitäten des täglichen Lebens selbst verrichten. In der Schweiz leben etwa 600 Personen mit ALS. Informell Pflegende, vorrangig Familienangehörige aber auch Freunde und Verwandte, die sogenannten *informal caregivers,* übernehmen eine zentrale Rolle bei der Begleitung, Betreuung und Pflege von Menschen mit ALS. Die Übernahme dieser Tätigkeiten kann für informell Pflegende belastend sein, sodass ein unterschiedlich gearteter Unterstützungsbedarf bestehen kann. Über den Unterstützungsbedarf dieser informell Pflegenden in der Schweiz ist wenig bekannt und auch international ist der Forschungsstand lückenhaft. Weiterhin ergeben sich für informell Pflegende und Menschen mit ALS als auch behandelnde Fachpersonen verschiedene ethische Herausforderungen.

Das erste Kapitel leitet in die Themen amyotrophe Lateralsklerose, Angehörigenpflege respektive informal caregiving, palliative und multidisziplinäre Care für Menschen mit ALS ein. Diese Einleitung bietet den Hintergrund für das Verständnis der einzelnen Kapitel.

Das zweite Kapitel legt Methoden, Forschungsfragen und Methodologie in empirischer Bioethik für die vorliegende Thesis dar. Um die verschiedenen Stränge dieser kumulativen Thesis zu verbinden, wird die Methodologie Critical Applied Ethics in diesem Kapitel vorgestellt.

Die Kapitel drei bis zehn bilden den Ergebnisteil dieser Arbeit, wobei ich zunächst einen Überblick über die Bedürfnisse von informell Pflegenden von Menschen mit ALS während des Krankheitsverlaufs in Form einer systematischen Übersichtsarbeit mittels narrativer Synthese darstelle. Diese Übersichtsarbeit synthetisiert Unterstützungsbedarf, der nach der Diagnose, während der Erkrankung und am Lebensende relevant ist. In Ergänzung zu dieser Synthese bietet das vierte Kapitel einen Überblick über den Unterstützungsbedarf von informell Pflegenden in der Schweiz anhand einer thematischen Analyse von Interviewdaten sowohl von informell

Pflegenden als auch von Gesundheitsfachpersonen. Das Kapitel skizziert verschiedenen Unterstützungsbedarf informeller Pflegender, wie z.B. die administrative Belastung, das Advance Care Planning im Kontext der ALS und unterstreicht die Bedeutung sozialer Unterstützung, sei es in Peer-Gruppen oder in sogenannten Caring Communities.

Ergänzend zu dieser weitgehend deskriptiven Forschung bietet das fünfte Kapitel einen grounded-theory-basierten Ansatz, um informelle Pflege während des Krankheits- und Pflegeverlaufs der ALS zu verstehen. Informelle Pflege wird als Aufrechterhaltung eines Drahtseilakts zwischen verschiedenen Aktivitäten verstanden, den informelle Pflegepersonen vollziehen.

In der systematischen Übersichtsarbeit haben wir eine Lücke in der Forschung zum Unterstützungsbedarf nach dem Tod gefunden. Daher bietet das sechste Kapitel eine thematische Analyse der Trauererfahrungen von informell Pflegenden. Das Kapitel diskutiert die Unterstützung nach dem Tod für informelle Pflegende von Menschen mit ALS im Rahmen eines etablierten Public-Health-Modells der Trauerbegleitung. Trauerbegleitung muss proaktiv von Fachleuten des Gesundheitswesens geleistet werden; sie muss jedoch bedarfsgerecht sein, um diejenigen anzusprechen, die sie benötigen und nicht diejenigen, die durch anderweitige Unterstützung bereits ausreichend unterstützt werden.

Vor dem Tod müssen Menschen mit ALS Entscheidungen bezüglich ihres Lebensendes treffen. Das siebte Kapitel befasst sich mit Entscheidungen am Lebensende mittels Brain-Computer-Interfaces. Ethische Herausforderungen, insbesondere im Hinblick auf die rechtliche Anforderung, die physische Kontrolle über das Pharmakon beim assistierten Suizid zu behalten, werden im Kontext der ALS dargestellt. Das Kapitel kommt zu dem Schluss, dass die physische Kontrolle die autonome Entscheidungsfindung bei ALS nicht einschränken sollte, wenn Menschen mit ALS über Brain-Computer-Interfaces kommunizieren und die Kontrolle ausüben können.

Das achte Kapitel skizziert informelle Pflege am Lebensende im häuslichen Bereich. Hier besteht bei der informellen Pflege am Lebensende in mehrfacher Hinsicht Unschärfe: zwischen einer informellen Pflegenden und einer nicht pflegenden nahestehenden Person, zwischen formellen bzw. professionellen und informellen Pflegepersonen und zwischen der Fürsorge für Andere und der Selbstfürsorge. Dieses

Kapitel ist nicht spezifisch für ALS sondern führt in die Feinheiten der informellen Pflege allgemein aus der Perspektive der Care Ethics ein.

Das neunte Kapitel diskutiert die Konzeptualisierung der Hoffnungslosigkeit im Kontext von ALS. Es wird oft angenommen, dass Hoffnungslosigkeit und Demoralisierung die Entscheidungsfähigkeit von Menschen mit ALS beeinträchtigen. Das Kapitel argumentiert, dass Hoffnungslosigkeit im Kontext der ALS jedoch oft falsch konzeptualisiert wird. Dieser Essay wurde mit dem Paul Wainwright Postgraduate Essay Prize 2019 ausgezeichnet.

Informell Pflegende benötigen für ihre Pflege unterschiedliche Unterstützung und Hilfsmittel. Das zehnte Kapitel identifiziert ungerechte Bedingungen bei der Bereitstellung von Assistenztechnologien und Hilfsmittel bei ALS, die durch eine Lücke im Übergang von der Invalidenversicherung zur Alters- und Hinterbliebenenversicherung in der Schweiz entstehen.

Die Thesis schließt mit einer kritischen Diskussion und Kontextualisierung der vorliegenden Forschung. Mithilfe der Critical Applied Ethics wird ein roter Faden gezogen, der die normativen und empirischen Dimensionen der Arbeit verbindet. Anschließend werden klinische und politische Empfehlungen vorgeschlagen, um die Situation der informellen Pflegenden von Menschen mit ALS zu verbessern.

Chapter 1.

Background

1. Background

The topic of this cumulative dissertation is informal caregiving for people with amyotrophic lateral sclerosis[1] (ALS) with an emphasis on palliative care. The dissertation was conducted within the PhD program of Biomedical Ethics at the University of Basel and financed by the Swiss Academy of Medical Sciences (SAMS; Palliative Care research program (PC 21/17)) under the title "*Palliative care at home: A multi perspective study on the complex needs of ALS family caregivers*".

Amyotrophic lateral sclerosis (ALS) is part of the motor neuron diseases but also synonymously known as motor neuron(e) disease (MND), Lou Gehrig's disease after a famous baseball player who developed ALS, or Charcot's disease because it was first described by neurologist Jean-Marie Charcot as a disease combination of different brain lesions. Charcot first described ALS in 1865 by separating "cases of acute-onset weakness from those with a slowly progressive course" (Goetz, 2000, p. 338).

ALS is a progressive neurodegenerative disease of the motor neurons in the brain and spinal cord leading to death from dyspnoea with a median life-expectancy of 2-4 years (Del Aguila et al., 2003) from first onset of symptoms. When breathing is not possible, mechanical ventilation via tracheostomy can prolong the life of patients for an average of 2 years (Niedermeyer et al., 2019).

As a terminal disease without a cure, care provided by healthcare providers for people with ALS encompasses both palliative and supportive care. Palliative care entails caring for the physical, emotional, and spiritual needs of the patient, according to the 2002 definition by the World Health Organization (Sepúlveda et al., 2002). Additionally, the 2002 definition of palliative care extended it to include care for family members of people with incurable diseases, as well as providing counsel to them before and after death. Nonetheless, family members are not only recipients of care by healthcare providers and other involved professionals, but they are also responsible for providing the largest part of care to the affected person with ALS (Aoun et al., 2013). In juxtaposition to professional, formal care, this care provided by informal caregivers is

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[1] In this thesis, the terms people (living) with ALS and patients with ALS might be used. They are, however, not interchangeable. People with ALS is a term used in a wider sense, encompassing all aspects of the lives lived with ALS, while the term patients is used when restricted to the healthcare context. Generally, the term people with ALS is seen as less ostracizing, and this pays tribute to the fact that most of care for people with ALS is indeed rendered outside of the healthcare context.

often called informal care (Zigante, 2018). Informal caregivers[2] are family members, friends, or acquaintances who do not act in a professional capacity. Besides this negative definition (informal caregiving as non-professional caregiving) and the aforementioned broad definition, there is a lack of standard definition of informal care in empirical research (Zigante, 2018). In this thesis, the broad definition is generally accepted and critically discussed with the blurred line to professional caregiving in Chapter 8. In the following, these central components of this thesis – ALS, multidisciplinary and palliative care, informal caregiving – will be introduced.

1.1 Amyotrophic Lateral Sclerosis

Clinical Presentation and Diagnosis of ALS

As a neurodegenerative disease primarily of the motor neurons, people with ALS present with motor deficits that can develop as quickly as a few weeks. Depending on the type of ALS, different voluntary muscle groups are affected and lead to different clinical presentations, often complicating an initial diagnosis (see below). In two thirds of patients, age of onset is between 50 and 70 years of age which is a stable pattern among time and cultures (Caroscio et al., 1987; Liu, Cui, & Fan, 2013). In light of demographic change in Europe, ALS will become significantly more frequent in ageing populations (Arthur et al., 2016). The prevalence of ALS in Europe and in people with European descent has been estimated at 2·2–3·0 cases per 100 000 people with lower incidences in other regions, e.g. East and South Asia (Logroscino & Piccinni, 2019), and with men being more commonly affected (Logroscino et al., 2010). Generally, the gender ratio of being diagnosed with ALS is between 1 to 3 from female to male patients (Manjaly et al., 2010).

ALS has been traditionally diagnosed, primarily for the purpose of clinical research, according to the El Escorial criteria (Brooks et al., 2000). These firstly developed criteria defined four categories of disease regarding the probability of disease: definite, probable, possible, and suspected ALS. The El Escorial criteria have been

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[2] The terminology of caregiver has also been subject to debate, as without doubt the caregivers of people with ALS are not solely caregivers, they are also wives, husbands, friends, lover, or children of people with ALS.

repeatedly revised to keep up with research as well as replaced by the Awaji criteria, though both criteria have been contested.

The last proposal of diagnostic criteria, both necessary and sufficient for the diagnosis ALS, from the year 2020 demands that there is progressive disorder of the motor system clinically present in at least one body region, but also gives note to cognitive and behavioural abnormalities (Shefner et al., 2020). While these new criteria rest on clinical foci, also a generalised symptom onset is recognized. Generally, in accordance with earlier criteria, affected clinical foci can be classified in accordance with the affected motor neuron regions as bulbar, cervical, thoracic, and lumbosacral (Shefner et al., 2020). However, for simplicity, types of ALS are often grouped into symptomatic site of onset: limb-onset (70% of patients) with upper and lower motor neurons symptoms in the limbs and bulbar-onset (25%) presenting with speech and swallowing symptoms (Kiernan et al., 2011). Common symptoms of limb-onset ALS are cramps, fasciculations, and muscle wasting in the limbs, in particular in the hands (Kiernan et al., 2011). The rest of patients present with initial respiratory or trunk symptoms or atypical symptoms. These are typically associated with poor prognosis. Motor neurons of eye and sphincter muscles are usually not affected. The latest diagnostic criteria include cognitive and behavioural criteria leading to a scientific debate on whether ALS and fronto-temporal dementia (FTD) are part of a continuum of disorder (Strong et al., 2017; Lulé et al., 2019).

About 5-10% of ALS is familial and several responsible genes have been identified (Kiernan et al., 2011), however these do not play a role for diagnosis of ALS. The other 90% of ALS cases are classified as sporadic cases with no known family history.

For the people and families affected by ALS, the search for a diagnosis starts often long before a correct diagnosis of ALS is provided. The average time period from symptom onset to diagnosis varies from many months to years. In a retrospective qualitative analysis with patients and caregivers, Househam and Swash (2000) found that in a cohort in England and Wales the mean time until diagnosis was 16.2 months. O'Brien and colleagues (2012) found that this often leads to a substandard diagnostic process leaving patients and caregivers unsatisfied. Cellura and colleagues (2012) studied the time to diagnosis and found that it differed according to diagnostic subtype, with limb-onset ALS being diagnosed after in median 12 months from symptom onset and two months later than bulbar-onset ALS. In a 20-year review, Mitchell and colleagues (2010) found the time relatively stable: “Time from first symptom to diagnosis hovered around 12 months, with the diagnosis typically being made around the midpoint (50% of total disease duration elapsed) of the disease pathway” (p. 537). While the authors urge to fast-track diagnostic processes, there is an inherent clinical complexity of ALS often rendering diagnosis difficult for primary care providers (Mitchell et al., 2010). Therefore, a reason for this diagnostic delay is the aforementioned diverse clinical presentation and speed of progression. However, Galvin and colleagues (2017) found that even if primary care providers directly refer patients to neurologists, this did not reduce diagnostic delay. In the same study, the authors estimated the cost of misdiagnosis and delayed diagnosis before patients enter multidisciplinary care at €3,486 per patient (Galvin et al., 2017).

An early diagnosis is therefore crucial for both patients as well as healthcare systems. Breaking the news and communication of diagnosis is intricate (Chio & Borasio, 2004). It is recommended that the diagnosis should be given by a skilled physician who knows the patient well in a private setting, allowing a comprehensive discussion of the implications of the diagnosis (Andersen et al., 2012). Care with communicating the diagnosis is important since patients often experience the diagnosis as a shock (Ozanne & Graneheim, 2017), and good communication is crucial to preserve the patient-physician relationship (Andersen et al., 2012).

Neuropsychological and behavioural sequalae of ALS

While 20 years ago, Borasio and colleagues (2001) wrote that “patients witness their progressive disability, due to degeneration of upper and lower motor neurons, with a fully clear mind” (p. 159), nowadays it is accepted that between 30%–50% of people with ALS have cognitive, emotional, and behavioural impairments (Giordana et al., 2010; Simon & Goldstein, 2018; Benbrika et al., 2019). Cognitive, emotional, and behavioural impairments are akin to the impairments seen in FTD. One of these is pathological laughing and crying, which is „characterized by episodes of involuntary outbursts of emotional expression” (Hübers et al., 2016, p. 1788). Furthermore, changes in social cognition and social behavior are present in up to a third of patients, which includes apathy, reduced empathy, and inflexibility. Especially apathy among the behavioural impairments is associated with increased caregiver burden (Caga et al., 2018). Up to half of patients present with cognitive impairment, however, this impairment is generally mild and affects only one cognitive domain; and only 5–15% present with symptoms akin to complete FTD (Lulé et al., 2019; Phukan et al., 2012; Goldstein & Abrahams, 2013). Due to these cognitive impairments, decision-making capacity of people with ALS has been called into question (see below in the subsection *Decision-making in ALS*).

1.2 Supportive and palliative care for people with ALS

Due to the complexity of symptoms and disease progression, several professional disciplines (e.g., physicians, nurses, occupational and physical therapists, psychologists, social workers) are involved in the care for people with ALS and their families. Multidisciplinary care is best provided and coordinated in specialised ALS/neuromuscular centres. This multidisciplinary care includes palliative care which improves quality of life of people with ALS (Hardiman & Phukan, 2009; Van den Berg et al., 2005; Ng, Khan, & Mathers, 2009), and possibly also survival (Ng, Khan, & Mathers, 2009; Paipa et al., 2019). While there is international consensus that multidisciplinary care and palliative care should be integrated early in caring for people with ALS and their families, the actual support offered differs in different healthcare systems (Bede et al., 2011). For example, multidisciplinary care in the Netherlands includes three different care coordinators during different disease stages while in the United Kingdom a linear pathway for end-of-life care has been developed (Bede et al., 2011).

Guidelines by the European Federation of Neurological Societies (EFNS; Andersen et al., 2012) outline that patients and families should be seen by the multidisciplinary care teams every 2-3 months, but more frequently if the disease progresses faster. Multidisciplinary care teams offer multiple lines of support, as displayed in Table 1.

Table 1: Multidisciplinary support adapted from Hogden et al., 2017

Situation	Support intervention/treatment	Multidisciplinary support
Disease progression	Riluzole	Neurologist/rehabilitation physician
Breathing difficulty	Assisted ventilation	Respiratory physician or therapist/Nursing
Eating and drinking difficulty	Gastrostomy	Gastroenterologist/Dietitian/Speech therapist/Nursing

Saliva management	Medication, Botox	Neurologist
Mobility	Mobility equipment	Physiotherapist/Occupational therapist/provider
Cognitive, behaviour and mood issues	Counselling and support for patients and families	Neuropsychologist, Psychologist
Self-care	Assistive equipment	Occupational therapist/Nursing/provider
Communication	Alternative communication devices	Speech therapists/Occupational therapist/provider
Grief and loss	Counselling and support for patients and families	Social worker/Psychologist/Palliative care team/provider
Carer support	Counselling and support for carers	Social worker/Palliative care team

The only disease-modifying drug available for people with ALS is Riluzole® (in Switzerland Rilutek®; Miller et al., 2009) which delays the progression of ALS in the later stages (Miller, Mitchell, & Moore, 2012; Hinchcliffe & Smith, 2017). Therefore, in the absence of a cure, progressive muscle weakness causes death by hypercapnia (Borasio & Voltz, 1998). As ALS remains an incurable disease, it is considered paradigmatic case for a disease of non-oncological palliative care research (Borasio et al., 2002).

While palliative physicians care for a wide range of medical issues in ALS, dyspnoea and pain are at the centre of care to uphold quality of life of people with ALS in close cooperation with ALS care specialist and other neurologists (Karam et al., 2016). Nonetheless, palliative care for this patient group goes beyond palliative medicine. It aims not only at symptom control but further at sustaining quality of life for both the patient as well as informal caregivers (Mitsumoto & Rabkin, 2007). Most of the support interventions are benign in their appliance in everyday life and should best be available before they are needed (assistive equipment, communication devices, among others). However, with progression of the disease increasingly interventions are needed to uphold quality of life and sustain life of people with ALS. This has effects on patients and family caregivers.

People with ALS often experience increasing psychological and existential distress during the disease course (Mitchell & Borasio, 2007). Psychosocial and spiritual care address this distress (Pagnini, 2013; Lambert, 2014). Generally, Meaning in Life has been a crucial concept in the palliative care (Fegg et al., 2010) and counselling is part of the multidisciplinary support offered in ALS centres. Existential decisions brought upon by increasing immobility, dyspnoea and progressive impairment demand particular attention in decision-making.

Decision-making in ALS

People with ALS and their families must make existential decisions during the disease course. As outlined in Table 1, these include gastrostomy, non-invasive and invasive ventilation, as well as assisted dying and euthanasia where these options are available. Due to internationally varied jurisdictions and multidisciplinary care, these decisions are rendered in different contexts but have major themes in common (Bede et al., 2011) Best practice and in line with international guidelines (Bede et al., 2011), these decisions should be discussed early and in a dialogical manner such as provided by advance care planning (ACP), and finally put into writing as an advance directive (Cipolletta & Reggiani, 2021). The process of ACP is routinely carried out in ALS centres nationally in Switzerland and internationally (Andersen et al., 2012; Sukockienė et al., 2021; Seeber et al., 2019), but can also be performed by general physicians (GPs; Bally, Krones, & Jox, 2020).

According to Seeber and colleagues (2019) ACP can be integrated into routine from diagnosis; however, timing of ACP is important, and reluctance can influence uptake of ACP (Sukockienė et al., 2021). Generally, ACP is known to improve quality of care and life of patients in palliative care by avoiding unwished interventions and decrease hospitalization (Brinkman-Stoppelenburg et al., 2014).

In general, physicians have the duty to provide information about the available treatment options within the framework of informed consent (Faden & Beauchamp, 1986; Lulé, Kübler, & Ludolph, 2019). Faden and Beauchamp (1986) famously distinguished two senses of informed consent: informed consent as autonomous authorization and informed consent as effective consent. Informed consent as autonomous authorization is "legally or institutionally effective" (Faden & Beauchamp, 1986, p. 280) consent. Informed consent as autonomous authorization is given "if a patient or subject with (1) substantial understanding and (2) in substantial absence of control by others (3) intentionally (4) authorizes a professional (to do I)" (Faden & Beauchamp, 1986, p. 278). Furthermore, informed consent is only valid if the person giving informed consent is legally competent to do so. Generally, informed consent therefore is constituted by five elements: “(1) competence, (2) disclosure, (3) understanding, (4) voluntariness, and (5) consent” (Beauchamp & Childress, 2001, p. 79). Most often, legal competence corresponds to clinical judgement of a physician on decision-making capacity of a person. In line with the bioethical principle of autonomy, patients with ALS need to have decision-making capacity for the consent to be valid. Doubt has been cast on the capacity of patients with ALS to render these decisions (Khin et al., 2015) due to the neuropsychiatric and cognitive impairments occurring in ALS (see above). However, capacity is always situational and therefore cannot be ruled out due to general cognitive impairment, as also in dementia generally (Poppe et al., 2020).

It is the role of the attending physician to provide judgement of capacity and enable decision-making in ALS. There is considerable influence of the physician on decision-making and more experienced physicians can support people with ALS and their families adequately (Aho-Özhan et al., 2017; Thurn et al., 2019). At the core of influence is the question of quality of life in ALS (Lulé, Kübler, & Ludolph, 2019). If physicians doubt the person with ALS’ well-being primarily due to physical impairment, this can lead them to influence decision-making against severe life-prolonging measures. Generally, this touches on what is widely known as the disability paradox (Ubel et al., 2005; Fellinghauer et al., 2012): that people with “severe impairments tend to report high quality of life” (Fellinghauer et al., 2012, p. 1) while physicians, especially those inexperienced with ALS, might conflate severe impairment with low quality of life (Aho-Özhan et al., 2017). However, even in the severely impaired state of locked-in state (LIS), people with ALS report high quality of life when being interviewed via eye-tracking control (Kuzma-Kozakiewicz et al., 2019).

Decision-making with regard to invasive ventilation and tracheostomy is different across cultures (Smyth et al., 1997), as is the case for end-of-life care in general (Gysels et al., 2012). There is evidence that invasive ventilation is discouraged in ALS in Switzerland and France, as it is generally in Europe (Heritier-Barras et al., 2013; Takei et al., 2017). This is likely due to the burden of care for informal caregivers and the lack of support structures in Switzerland for ventilated patients (see Chapter 4). In contrast, invasive ventilation is used more frequently in Japan. Almost a third of patients with ALS choose invasive ventilation to prolong their lives (Takei et al., 2017; Hirano & Yamazaki, 2010). There are several reasons for the higher uptake of ventilation in Japan.

According to Hirano and Yamazaki (2010), they comprise “more permissive social insurance coverage, physician paternalism, and clinical guidelines recommending that physicians consult both patients and their families” (p. 51). In the same vein, access to inexpensive long-term ventilation at home is highlighted as a critical factor in favour of tracheostomy (Kawaguchi, 2019). The examples of Switzerland and Japan point to the importance of contextual factors in decision-making of invasive ventilation in ALS.

In case of absence of capacity or inability to communicate wishes and in the absence of advance care planning and living wills, family members who are often also informal caregivers are next in the cascade of surrogate decision-making (Buchanan & Brock, 1989; Krones, 2016).

1.3 Informal caregiving in ALS

Due to the loss of independence in daily living, people with ALS rely on the support and care of others. Most of this support is rendered by informal caregivers and increases with the progression of ALS. These informal caregivers thus not only have to acquire a range of skills relating to the physical and medical needs of the patients (Rabkin & Albert, 2009; Murphy et al., 2009), but they also have to coordinate support services and organize finances (Mockford, Jenkison, & Fitzpatrick, 2006; Gladman & Zinman, 2015), among others. Due to the increasing responsibilities, as well as the emotional toll of facing a fatal disease and uncertain future, informal caregivers are strained by caregiving and experience caregiver burden. Caregiver burden entails emotional, physical, social, and financial burden (de Wit et al., 2018). International research has hence mainly focused on quality of life and burden of care of informal caregivers of patients with ALS. It is reported that the objective burden of care increases with impairment in physical functioning of the patient (de Wit et al., 2018). Likely, this is due to the higher immobility of the patient and the necessity of more medical treatment and support interventions. However, the subjectively experienced burden of care by informal caregivers is only indirectly mediated by the higher immobility of the person with ALS (Hecht et al., 2003).

Caregiver burden is also associated with lower perceived control over caregiving (de Wit et al., 2020) and caregiver distress and subjective burden of care are more affected by the cognitive and behavioural changes of the patient (Burke et al., 2015, 2017), e.g., pseudobulbar affect or frontotemporal dementia (Merrilees et al., 2010). Furthermore, depression of the caregiver influences the subjectively perceived burden of care (de Wit et al., 2018). Nonetheless, support needs of informal caregivers are more encompassing than management of these issues. In fact, one of the literature reviews concluded that

> "while caregiver burden and QOL studies have dominated the research and documented the substantial distress experienced by MND family caregivers, more studies are now emerging that provide guidance about how to provide optimal palliative and supportive care to people with MND and their family caregivers" (Aoun et al., 2013, p. 442).

Hence, as it is not easily possible to infer support needs of informal caregivers from these caregiver burden and quality-of-life studies, health services researchers in the context of ALS have started to investigate conditions of informal caregiving. For example, an Irish study of informal caregivers' needs from first onset of symptoms to bereavement (Galvin et al., 2016; 2017) emphasized the need for taking the different trajectories of informal caregiving for people with ALS into account. In general, this means that different needs occur at the onset of symptoms than at the later stages of (palliative) informal caregiving and that provision of healthcare services professionals should be adapted to different needs of caregivers at these stages (Galvin et al., 2017). In a Dutch qualitative study, the authors assessed ALS caregivers' needs (de Wit et al., 2018) before developing further quantitative approaches to evaluate the burden of caregiving (de Wit et al., 2018). In their qualitative study, they described caregiving in ALS as "an intensive caregiving situation with multiple needs emerging in a short period" (de Wit, 2018, p. 1) among them, more personal time, assistance, counselling, and peer contact. Multiple needs encompass training in physical care of the patient and handling of appliances, among others (O'Brien et al., 2012). Further needs of informal caregivers occur in inpatient care during hospital stays of the patient (Larsson et al., 2015). Naturally, these forms of qualitative research are not representative for the needs of all informal caregivers of people with ALS in these countries, but they show the nationally different specific conditions of caregiving in the context of ALS.

In Switzerland, little is known about the support needs of informal caregivers of people with ALS. The existing Swiss research has focused on wishes-for-hastened-death and attitudes towards assisted suicide among people with ALS (Stutzki et al., 2012; Stutzki et al., 2014) and their informal caregivers; or on impact of other specific medical interventions e.g., tracheostomy (Heritier-Barras et al., 2013) on the family caregiver. The Swiss position of having both regimes of palliative care and assisted suicide as possible end-of-life options is relevant to informal caregivers who often must support their spouses or family members in their end-of-life option. In contrast to the dearth of research on informal caregivers' needs, basic and applied medical research has been conducted within Switzerland especially on biomarkers and neurodegenerative processes of ALS (e.g., Czell et al., 2013).

Given the lack of research on informal caregivers' needs, the scope of the research presented in this thesis was deliberately kept open. It follows that a variety of domains are covered ranging from practical training needs (how to handle medical technology and physical care work, i.e., transfers and daily hygiene) to ethical needs relating to shared decision-making and advanced care planning at the later stages. This thesis therefore provides an explorative picture of informal caregiving in supportive and palliative care for people with ALS.

1.5 References

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Chapter 2.

Research Objectives, Methods, and Methodology

2.1 Research questions and objectives

The overall goal of this thesis is to provide a *Lay the Land* (Kon, 2009) of the multidimensional needs of informal caregivers to understand their unmet needs and ultimately allow future research and needs-adapted interventions to ameliorate their situation. In turn, addressing unmet needs of informal caregivers should also improve the conditions of palliative care of people with ALS. Furthermore, such knowledge is critical for people with ALS as the support available to them through informal caregivers and healthcare professionals is an important factor to determine the preferred end-of-life circumstances.

The research questions to address the overall goal of the thesis are:

> ***A***. *What is the synthesized evidence of supportive needs of informal caregivers of patients with ALS across the caregiving course?*
>
> *B. What are the supportive needs of informal caregivers of people with ALS in Switzerland?*
>
> ***C***. *How can we understand informal caregiving during palliative care for people with ALS in particular?*
>
> ***D***. *What ethical, legal, and social issues (ELSI) are there at the end of life for people with ALS and their caregivers?*

From these research questions, the following research objectives can be derived.

Research objectives addressing these questions

A. What is the synthesized evidence of supportive needs of informal caregivers of patients with ALS across the caregiving course?

The first objective of this thesis is to provide a systematic review of the existing evidence of ALS informal caregivers' needs at different stages of caregiving. Former literature reviews (Mockford, Jenkinson, & Fitzpatrick, 2006; Aoun et al., 2013; Oh & Kim, 2017) have been unsystematic, i.e., these reviews did not assess the quality of evidence. Furthermore, these earlier literature reviews included almost all available literature on informal caregivers of patients with ALS (e.g., also quality of life and burden of care studies) and on patients with ALS, probably due to the general small size of the field of research. In contrast, the systematic review in this thesis extracts specific needs of informal caregivers and assesses the evidence with quality criteria for quantitative and qualitative designs. Additionally, these needs are linked to different stages of caregiving (Williams et al., 2008), which is vitally needed for any guidance in practice and further research.

B. What are the supportive needs of informal caregivers of people with ALS in Switzerland?
The second research objective is to provide insights into Swiss ALS informal caregivers needs across the caregiving course. As laid out in Chapter 1, there is a lack of research on needs of informal caregivers of people with ALS which is needed for the provision of palliative care in Switzerland. Furthermore, exploration of informal caregivers' needs at the end of life of the person with ALS and after bereavement are a largely unexplored field of research.

C. How can we understand informal caregiving during palliative care for people with ALS?
The third objective of this thesis is to add to the international research on caregivers need by providing a theory of caregiving at the end of life (and after bereavement). Caregiving in the context of chronic diseases has been described as 'trajectory work' (Corbin & Strauss, 1985, p. 238), subsuming three different lines of work: (1) illness work, (2) everyday life work, and (3) biographical work. Thus, the concept of work here goes beyond managing (everyday) tasks. While Corbin and Strauss have described caregiving work during the caregiving trajectory for chronic disease, there is lack of research for fatal diseases, especially concerning the terminal phase and bereavement of caregivers.

D. What ethical, legal, and social issues (ELSI) are there at the end of life for people with ALS and their caregivers?
The primary focus of this thesis is to provide empirical data on supportive needs of informal caregivers. However, the interviews and systematic review also revealed a range of ethical, legal, and social issues (ELSI) people with ALS and their informal caregivers were confronted with. Hence, research objectives A—C are complemented by conceptual analysis of ELSI. These research objectives are addressed by the following methods.

2.2 Methods

This thesis employs four different types of methods: a) systematic review using narrative synthesis, b) qualitative interview research using thematic analysis, c) qualitative interview research using a grounded theory approach, and d) conceptual analysis.

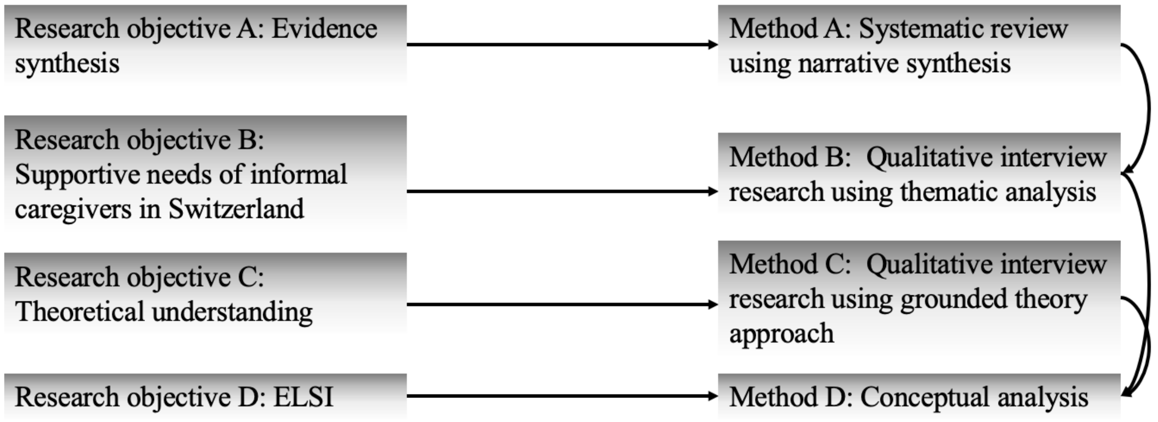

Figure 1: Methods to match the Research Objectives

Method A: Systematic review

The systematic review (SR) on ALS caregiver needs throughout the caregiving trajectory adheres to the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA; Moher et al., 2009) guidelines. Additionally, the systematic review is based on an adapted PICO (PCO; population, context, outcome) approach. Since most studies in the field of ALS caregiver research are non-randomized quantitative and qualitative studies, both quality assessment and analysis strategy had to be adapted to meet the objective of the SR. Quality assessment for qualitative studies used the checklist for qualitative research from the Joanna-Briggs-Institute (Hannes, Lockwood, & Pearson, 2010), while the quality of quantitative (mostly descriptive) studies was assessed using a modified quality assessment tool for observational cohort and cross-sectional studies by the National Institute of Health (NIH). The extraction sheet can be found in Appendix D. Data analysis was based on narrative synthesis which is justified given the different designs of the studies included (Popay et al., 2006). Further details on search strategy, data extraction, and data analysis can be found in the pre-registered PROSPERO protocol in Appendix C. Based on this SR, interview guides for the semi-structured interviews were developed.

Methods B and C: Study design of the interview study

The empirical study is based on in-depth semi-structured interviews with informal caregivers of people with ALS and healthcare professionals conducted in the years 2018—2020. In cross-sectional interviews, two groups of informal caregivers were interviewed: current informal caregivers and bereaved informal caregivers of people with ALS. Additionally, interviews with healthcare professionals (nurses, physicians, therapists) were conducted. In triangulating data from these groups, both the current needs expressed by informal caregivers as well as needs in retrospective are accessible, as well as the external perspective of healthcare professionals. Furthermore, the period of death and bereavement in this way can be covered from both prospective as well as retrospective angles allowing us to access problems which might not be expected by informal caregivers beforehand. The literature-based (see above) and pilot-tested interview guide is semi-structured with an opening narrative part (Mey & Mruck, 2007) and probing questions to ascertain the interviewer covers possible domains of needs (e.g, informational, practical, organizational, physical care, advance care planning). The interview guide (in the English translation) can be found in the Appendix B.

All interviews were recorded as digital audio and transcribed using MaxQDA (VERBI, 2020). While transcribing, all identifying characteristics of participants were pseudonymized.

Sample

The sample consisted of 9 current caregivers, 14 bereaved informal caregivers, and 13 healthcare professionals (physicians, physiotherapists, nurses) providing care to individuals with ALS and their families. These numbers are justified by feasibility within the research project and are above a commonly accepted size in qualitative research (Mason, 2010). Informal caregivers were mostly family caregivers, with an age range from 28 to 74 years. From the family caregivers, most (96 %) were spouses or daughters of individuals diagnosed with ALS. Of the 23, nine informal caregivers were currently providing care to a person with ALS. Informal caregivers were recruited purposefully through ALS centres in German speaking part of Switzerland, through patient initiatives (Verein ALS Schweiz), peer contacts of former caregivers, and snowball sampling.

Especially notable help with recruitment was provided by Dr. Kathi Schweikert (REHAB Basel) and by Prof. Dr. Markus Weber (Muskelzentrum St. Gallen).

Informal caregivers contacted the interviewers via phone, mail, or email after having heard of the study. With informal caregivers, interviews took place mainly in person in the participants' home to accommodate time and caregiving constraints. Healthcare professionals were mostly contacted in advance and interviewed via telephone. Interviews were conducted and audiotaped after all participants had provided written informed consent. Interviews were transcribed verbatim in Standard High German using MAXQDA (Verbi Software, 2020).

The interviewers were a female physician (Luzia Iseli) working in general practice and a male clinical psychologist (the thesis author). Both had prior practice in conducting qualitative research interviews or analysis and were trained in qualitative data collection. The first four interviews served as a pilot for the semi-structured interview guide and the interviewers' interview skills, which were supervised by a senior researcher. For informal caregivers, the interview guide comprised of questions regarding practical supportive needs during caregiving, difficult situations, advance care planning and end-of-life, as well as the time after bereavement (see Appendix B). For healthcare professionals, the interview guide centred on support for caregivers and their own support (see Appendix B). The interviewers did not have any prior relationship to the research participants, nor did the participants have specific knowledge about the interviewers apart from general information about the project.

Coding and Analysis

For both qualitative research methods, the first step was inductively coding segment by segment of the interview transcription. Segments consisted of units that relayed a meaning relevant for the context (usually one to three sentences). Data analysis was performed in parallel to data collection. Data saturation was monitored while inductively coding and achieved when recurrent coding occurred. Data saturation was therefore understood as post-hoc information redundancy achieved during data analysis (Braun & Clarke, 2019). Overall, 20 interview transcripts were coded by hand by teams of two to three coders (six coders overall) to satisfy inter-coder agreement of approach to the data. The remaining interviews were coded inductively using MaxQDA solely by the first author. Generally, coding was an iterative process switching between codes and categories for an accurate understanding.

Method B: Thematic analysis

The exploratory part of the thesis uses thematic analysis (Braun & Clarke, 2006). Thematic analysis offers a flexible framework for different research objectives. It fits several research methodologies and epistemologies (Braun & Clarke, 2006, p. 97). As laid out by Braun and Clarke (2006), thematic analysis usually follows six phases of analysis (see Table 1).

Table 1: Phases in thematic analysis (Braun & Clarke, 2006, p. 87)
1. Familiarising yourself with your data
2. Generating initial codes
3. Searching for themes
4. Reviewing themes
5. Defining and naming themes
6. Producing the report

In following these phases, we arrived at themes of support needs of informal caregivers of people with ALS (Chapter 4 and 6).

Method C: Grounded theory-based approach

The qualitative research methodology based on grounded theory is commonly associated with research on caregiving work and trajectories in chronic disease (Corbin & Strauss, 1985) but is adapted here to develop a theoretical understanding of informal caregiving in palliative and end-of-life care for people in ALS. This research methodology is needed to meet the third objective of this thesis and to provide a theoretical understanding grounded in the empirical data. Nonetheless, this grounded theory-based approach is not only based empirical data, but also on theoretical presuppositions (Glaser, 1998; Charmaz, 2006).

As outlined above, the present research project employs purposive and snowball sampling due to practical constraints. This is a deviation from the traditional research program of Grounded Theory where theoretical sampling a defining feature (Glaser, 1998; Charmaz, 2006). Theoretical sampling conventionally is understood as new recruitments of participants in accordance with emergence of a theory, e.g., understanding informal caregiving within different relationship dynamics (friend versus spousal caregiver). As a grounded theory-based approach can lead to a better understanding of the process of informal caregiving, it can be used to delineate policy guidance to inform the relevant stakeholders on the needs of caregivers.

Method D: Conceptual analysis

Methods in normative ethics are notoriously hard to pinpoint, and so is the broadly termed method of conceptual analysis (Burgess, Cappelen, & Plunkett, 2020). Broadly, conceptual analysis in this thesis can be understood as an explication of conditions and conceptual background to a specific normative issue. The starting point of conceptual analysis were normative issues mentioned in the theoretical literature or raised by informal caregivers in the interviews. Chapter 7 and 8 deal with end-of-life decision-making with brain-computer interfaces in ALS and blurred boundaries of informal caregiving, for example when informal caregivers are employed by the state. Chapter 9 deals with an adequate understanding of hopelessness in ALS starting from theoretical considerations. Chapter 10 argues that there are gaps in fair access to assistive technology in social insurances in Switzerland. Most of these issues were raised by informal caregivers in the interviews. However, these conceptual analyses explicate issues on a normative level without using extensive empirical research.

2.3 Methodology in Empirical Bioethics

As stated previously, the thesis is primarily the result of the qualitative research conducted in the umbrella research project "*Palliative care at home: A multi perspective study on the complex needs of ALS family caregivers*" funded by the Swiss Academy of Medical Sciences (SAMS; Grant PC 21/07). As palliative care is concerned with existential decisions (e.g., withdrawing or refusing nutritional or respiratory life-sustaining treatments), normative considerations play a crucial role in the care for people with ALS (Daniel-Brunaud et al., 2017). Furthermore, informal care by itself is determined by social and moral norms. Additionally, also formally provided nursing care has been described as consisting of a moral attitude (Gastmans, 1999). From the perspective of Ethics of Care, this moral attitude is part of the *caring for—caring about* dichotomy often described as part of care (Cronqvist et al., 2004). It is therefore necessary to lay out a methodology that allows to combine normative and empirical research that is carried out in the final chapter of this thesis.

Biomedical ethics is concerned with moral norms in biomedical disciplines. Research on moral norms can be normative (what moral norms should be) and descriptive (what moral norms commonly are). From its inception, biomedical ethics have been concerned with facts. For normative bioethics, we need to know what the facts are that the moral norms should guide. However, the relationships between norms, what should be, and facts, what is, has been philosophically contested for at least 240 years since Hume's third treatise. In his famous paragraph on the Is-Ought gap (Hume, 1739 cited after Selby-Bigge, 1896, p. 469):

> In every system of morality, which I have hitherto met with, I have always remark'd, that the author proceeds for some time in the ordinary way of reasoning, and establishes the being of a God, or makes observations concerning human affairs; when of a sudden I am surpriz'd to find, that instead of the usual copulations of propositions, *is*, and *is not*, I meet with no proposition that is not connected with an *ought*, or an *ought not*. This change is imperceptible; but is, however, of the last consequence, for as this *ought*, or *ought not*, expresses some new relation or affirmation, 'tis necessary that it shou'd be observ'd and explain'd; and at the same time that a reason should be given, for what seems altogether inconceivable, how this new relation can be a deduction from others, which are entirely different from it.

Bioethics operates at this intersection, and therefore has been concerned at the heart of its methodology with the integration of fact and values, descriptions and prescriptions, or, simply, is and ought.

Overview of methodological integration in empirical bioethics

In the last 20 years, there has been an increased interest in the methodology of empirical bioethics to integrate normative considerations and empirical data (Wangmo & Provost, 2017; Wangmo et al., 2018). The incorporation of empirical data in research in bioethics has grown rapidly. This can be attributed to the interdisciplinary nature of bioethics on the one hand, but also to the experience of clinical ethicists that a top-down approach from moral principles is often not appropriate in clinical situations.

Indeed, there have been numerous methodological attempts to combine facts with moral norms, so many, that some authors have started to ask, "why count the ways?" of empirical research in bioethics (Sugarman, Kass, & Faden, 2009).

Given the centrality of informal care in the thesis, Care Ethics/Ethics of Care (EoC) necessarily provide a normative starting point to this thesis. Ethics of Care "start from the notion that humans are intrinsically dependent on and interconnected with one another" (Stensöta, 2010, p. 297). EoC especially has a long tradition of embedding care within empirical work and bridging dimensions of care into the public life (Tronto, 1993; Kitttay, 2009). According to Eva Feder Kittay, EoC starts from "actual activities and interactions and understands norms to be embedded in the actual" (Kittay, 2009, p. 123). Comparing several forms of care, we would pick "the best practices being minimally those in which each individual who is affected by the activities constituting the practice is not adversely affected and receives the maximum value of that practice, compatible with every other such individual getting the maximum value" (Kittay, 2009, p. 126-127). From this perspective on EoC, using this decision rule we could therefore determine the best practices of care for informal caregivers. However, while this decision rule is helpful to determine the value of a practice for an individual, it does not constitute a comprehensive framework combining normative and empirical considerations which can be applied in this thesis. For the purpose of this thesis, a more refined process of picking, evaluating empirical practices and their directing norms is necessary. Therefore, in the next section, different approaches to empirical bioethics will be shortly presented.

Overview of bioethical methodologies

Research strategies in empirical bioethics can be classified in several ways. In a systematic review of bioethical research strategies, Davies, Ives, and Dunn (2015) found that the most part of empirical research in bioethics combines normative research either in a dialogical or in a consultative manner. Briefly put, dialogical research invokes more the stakeholders' opinion about the ethical situation whilst consultative research consults with them but draws normative conclusions apart from them. Consultative bioethical research can be again divided into reflective equilibrium-based research (RE) and non-reflective equilibrium (non-RE) based approaches. Citing De Vries (2004), the authors differentiate four different empirical research strategies that underly all approaches in empirical bioethics (Davies, Ives, & Dunn, 2015, p. 2):

> (i) use empirical data to describe attitudes toward an issue;
>
> (ii) use empirical data to explore the likely or actual consequences of bioethical policies and decisions;
>
> (iii) use empirical data to explore the 'implicit normativity' in scientific/clinical practice, and
>
> (iv) use empirical data to understand the institution of bioethics.

Similarly, Alexander Kon (2009) classified empirical research in biomedical ethics into four categories: *Lay of the Land, Ideal versus Reality, Improving Care*, and *Changing Ethical Norms.* Bioethical studies within the category of *Lay of the Land* „seek to define current practices, opinions, beliefs, or other aspects that may be considered the status quo" (Kon, 2009, p. 60), while *Ideal versus Reality* assesses how far a normative ideal is represented in this status quo (say autonomy as informed consent in everyday clinical practice). *Improving Care* is interventional research addressing failures of the status quo to conform to normative ideals, in Kon's typology. Lastly, *Changing Ethical Norms* builds on top of the three types and based on them recommends the changing of a normative ideal. Kon's (2009) typology is very useful due to its simplicity, however, it remains undifferentiated. Another different typology is given by Molewijk and colleagues (2004, cited after Davies, Ives, and Dunn, 2015, p. 2). In their typology, empirical research can be situated in relation to moral theory to:

> [(a)] give complete authority to moral theory, and only use empirical data to provide evidence for premises or support factual claims;
>
> [(b)] give precedence to moral theory but accommodate a one-way relationship between theory and data such that empirical research can be used to refine theory;
>
> [(c)] give equal authority to both theory and data, such that both theory and interpretation of data can be adjusted in light of the other, and:
>
> [(d)] remove theory altogether from ethical analysis and focus only on the particulars, which are identified through empirical research.

In short, the researchers in these fields have been named the *Prescriptive Applied Ethicists* (a), the *Theorists* (b), *Critical Applied Ethicists* (c), and the *Particularists* (d). Due to its focus on both theory and empirical research, Critical Applied Ethics (CAE) stands out as the most suitable methodology for the presented research. CAE belongs to the consultative, non-RE based approached to empirical bioethics. Consultative here means that empirical data (of stakeholders' views) plays an important part in ethics research but is not sufficient for the normative conclusions. The latter is still carried out by the researcher or the "thinker" (Davies, Ives, and Dunn, 2015, p. 9). Non-RE describes an integrative approach to empirical and normative research than balancing different beliefs or facts about living conditions to gain coherence in moral theory. CAE according to Leget, Borry, and De Vries (2009) takes a five-step approach to these issues, and every step includes both a normative and an empirical dimension.

Firstly, the problem is determined. This can be done either from the normative or from the empirical entry-point. While most times problems are determined from the normative angle (e.g., violation of moral or legal principles), the authors note that this is prone to be biased by the ethicist's own focus on "more esoteric problems and ethical puzzles that are interesting to them" (Leget, Borry, & De Vries, 2009, p. 232).

Secondly, the problem is described. On the theoretical level, involved concepts are explicated (e.g., dignity, autonomy). The empirical side deals with motivations of involved agents (e.g., informal caregivers and people with ALS, nurses). The theoretical ethicists also shed light on evaluative descriptions in empirical research, while the empirical ethicists point out social context of decision-making and context for concepts.

Thirdly, effects and alternatives are pondered. This is the consequentialist aspect of CAE, where consequences of a decision/policy are empirically appraised. One good example in normative ethics is the 'slippery slope' argument (Williams, 1995). A slippery slope argument says that if a certain decision/policy is allowed then there is a natural progression towards another, possibly horrible, result (Williams, 1995). Hence, in this step, the effects of a decision/policy can be tested to see whether a slippery slope indeed leads to this result. Furthermore, alternatives for a policy can be found by interviewing experts or stakeholder in the field.

In the fourth step, the empirical data is normatively weighed. This step makes the process consultative: the empirical data has no more say in it. This process, according to Leget and colleagues (2009), should be done transparently. That means that one should be frank about one's own normative commitments and social positionality, a claim now shared among many bioethicists (Salloch et al., 2015) and also in corresponding qualitative methods (Berger, 2015). In the fifth and final step, the effect of a decision is evaluated which fits the description of Kon's (2009) categories *Improving Care* and *Changing Ethical Norms*.

Methodology in this thesis

The formal process of CAE is by itself a normative ideal for empirical research in bioethics. Due to practical constraints of the research project, it could only be applied post-hoc in this thesis. Using CAE, the different results from the four methods of this thesis will be combined and interlinked in the discussion (Chapter 11). By combining the different results of this thesis, a mosaic overview of informal caregiving in ALS from the perspective of empirical bioethics will be presented. In light of the primarily empirical nature of research presented in this thesis, most of the research necessarily falls in the category of identification and description of the problems (step one and two). However, the five steps of CAE show the relation between the results of different methods as well as gaps which exist due to the cumulative nature of this thesis.

2.4 Ethical considerations and institutional review

Ethical considerations relate to the risk for the participants involved in the empirical study, mainly emotional burden for current and bereaved caregivers for participating in the interview. The interviewers were sensitized for these issues with the possibility for research participants to take a break from the interview or to end it. In general, studies indicate that the participation in qualitative interviews for palliative care patients and caregivers can be helpful and can have empowering effects (Gysels, Shipman & Higginson, 2008).

A full research ethics application for the umbrella research project has been submitted in July 2018 and the cantonal research ethics committee (Ethikkommission Nordwest- und Zentralschweiz; EKNZ) decided that the research does not need full ethical review. Hence, a jurisdictional inquiry by the EKNZ waived ethics approval for this research in accordance with Swiss laws (see Appendix A). However, written informed consent was ascertained from every participant.

2.5. Peer-reviewed publications included as chapters in the results

As this is a cumulative thesis, the chapters included in the results are submitted or published peer-reviewed publications.

- **Chapter 3. Differentiating needs of informal caregivers of individuals with ALS across the caregiving course: a systematic review.**

  Poppe, C., Koné, I., Iseli, L. M., Schweikert, K., Elger, B. S., & Wangmo, T. (2020). Differentiating needs of informal caregivers of individuals with ALS across the caregiving course: a systematic review. *Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration*, 21(7-8), 519-541. https://doi.org/10.1080/21678421.2020.1771735

  Contribution: review protocol concept and design, systematic search, full-text review, abstract screening, data extraction, data analyses, manuscript writing.
- **Chapter 4. Supportive needs of informal caregivers of people with amyotrophic lateral sclerosis in Switzerland: A qualitative study.**

  Poppe, C., Schweikert, K., Krones, T., & Wangmo, T. (2022). Supportive needs of informal caregivers of people with amyotrophic lateral sclerosis in Switzerland: A qualitative study. Palliative Care and Social Practice, 16: 1–14, https://doi.org/10.1177/26323524221077700

  Contribution: research study design, data collection and transcription, coding, qualitative data analysis using thematic analysis, manuscript writing.
- **Chapter 5. “Walking a tight rope”: A qualitative study on informal caregiving in palliative care for amyotrophic lateral sclerosis.**

  Poppe, C., Verwey, M. & Wangmo, T. (2021). "Walking a tightrope": A grounded theory approach to informal caregiving for amyotrophic lateral sclerosis. *Health and Social Care in the Community*, 30(5), e1935-e1947. *https://doi.org/10.1111/hsc.13625*

  Contribution: research study design, data collection and transcription, coding, qualitative analysis using a grounded-theory approach, manuscript writing
- **Chapter 6. Bereavement and support experiences of informal caregivers of persons with amyotrophic lateral sclerosis: A qualitative study.**

Poppe, C., Iseli, L., Verwey, M., Wangmo, T. (2022). Bereavement and support experiences of informal caregivers of persons with amyotrophic lateral sclerosis: A qualitative study. *Journal of Social Work in End-of-Life & Palliative Care*, 18(1), 63-79. https://doi.org/10.1080/15524256.2021.1976352

Contribution: research study design, data collection and transcription, coding, qualitative data analysis using thematic analysis, writing manuscript

- **Chapter 7. Brain-computer-interfaces, completely locked-in state, and end-of-life decisions**

  Poppe, C. & Elger, B.S. (accepted). Brain-computer-interfaces, completely locked-in state in neurodegenerative diseases, and end-of-life decisions. *Journal of Bioethical Inquiry.*

  Contribution: literature review, conceptual analysis, manuscript writing
- **Chapter 8. Blurring Boundaries at the End of Life in Home Care.**

  Kaiser, M.*, Kuhn, E.*, Poppe, C.*, & Voss, H.* (2022). Blurring Boundaries at the End of Life in Home Care: A Look at Germany, Switzerland and the United Kingdom in the Light of Care Ethics. In: Salloch, S. & Seidlein, A.-H. (2022) Professional Ethics at the End of Life: European Perspectives. Philosophy and Medicine, Volume 141. Springer.

  *Equal first authorship

  Contribution: literature review, conceptual analysis of a part of the chapter, manuscript writing, revising manuscript
- **Chapter 9. Hopelessness in palliative care for people with motor neurone disease: Conceptual considerations.**

  Poppe, C. (2020). Hopelessness in palliative care for people with motor neurone disease: Conceptual considerations. *Nursing Ethics*, 27(1), 316–320. https://doi.org/10.1177/0969733019901225

  Contribution: literature review, conceptual analysis, manuscript writing
- **Chapter 10. Fair access to assistive technology? Gaps in transition from invalidity to old-age insurance in Switzerland.**

  Poppe, C., Martani, A., Schweikert, S. & Wangmo, T. (2022). Fair access to assistive technology? Gaps in transition from invalidity to old-age insurance in

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Contribution: literature review, conceptual analysis, manuscript writing

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Chapter 3.

Differentiating needs of informal caregivers of individuals with ALS across the caregiving course: a systematic review

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Differentiating needs of informal caregivers of individuals with ALS across the caregiving course: a systematic review

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Abstract

Background: Informal caregivers of people with amyotrophic lateral sclerosis (ALS) experience a range of needs across the course of the disease. For the provision of adequate support, an examination of the empirical evidence is necessary.

Aim: The purpose of the systematic review was to synthesize evidence of needs of informal caregivers of people with ALS at different stages of caregiving.

Method: Systematic review of empirical research on needs of ALS informal caregivers in both English and German, from January 2000 to August 2018. We searched the databases EMBASE, MEDLINE (PubMed), PsycINFO and CINAHL. Study selection, quality assessment, and data extraction was performed independently. Both quantitative and qualitative studies were included. Of the included studies, we additionally screened citing literature in Google Scholar (citation tracking). We linked the narrative synthesis to four stages of caregiving described by Williams and colleagues

(Williams et al., 2008) and used descriptive inductive thematic analysis to structure data within the stages.

Results: From 3275 abstracts screened, 48 manuscripts met our inclusion criteria. Our data analysis shows that needs differ across the four caregiving stages. While the stage of bereavement (4.) includes too little data for separate themes, themes for needs after diagnosis (1.), and terminal stage (3.) could be specified. As the maintenance (2.) stage comprised of themes relevant across the caregiving course, it became an overall stage.

Discussion: Healthcare professionals need to pay attention to current caregiving stages to provide support for informal caregivers. Further research is needed to tease out support needs for the bereavement phase.

Keywords

Informal caregivers, needs, support, disease course, caregiving stages

Preregistration

A protocol was preregistered on PROSPERO (CRD42018105311).

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1. Introduction

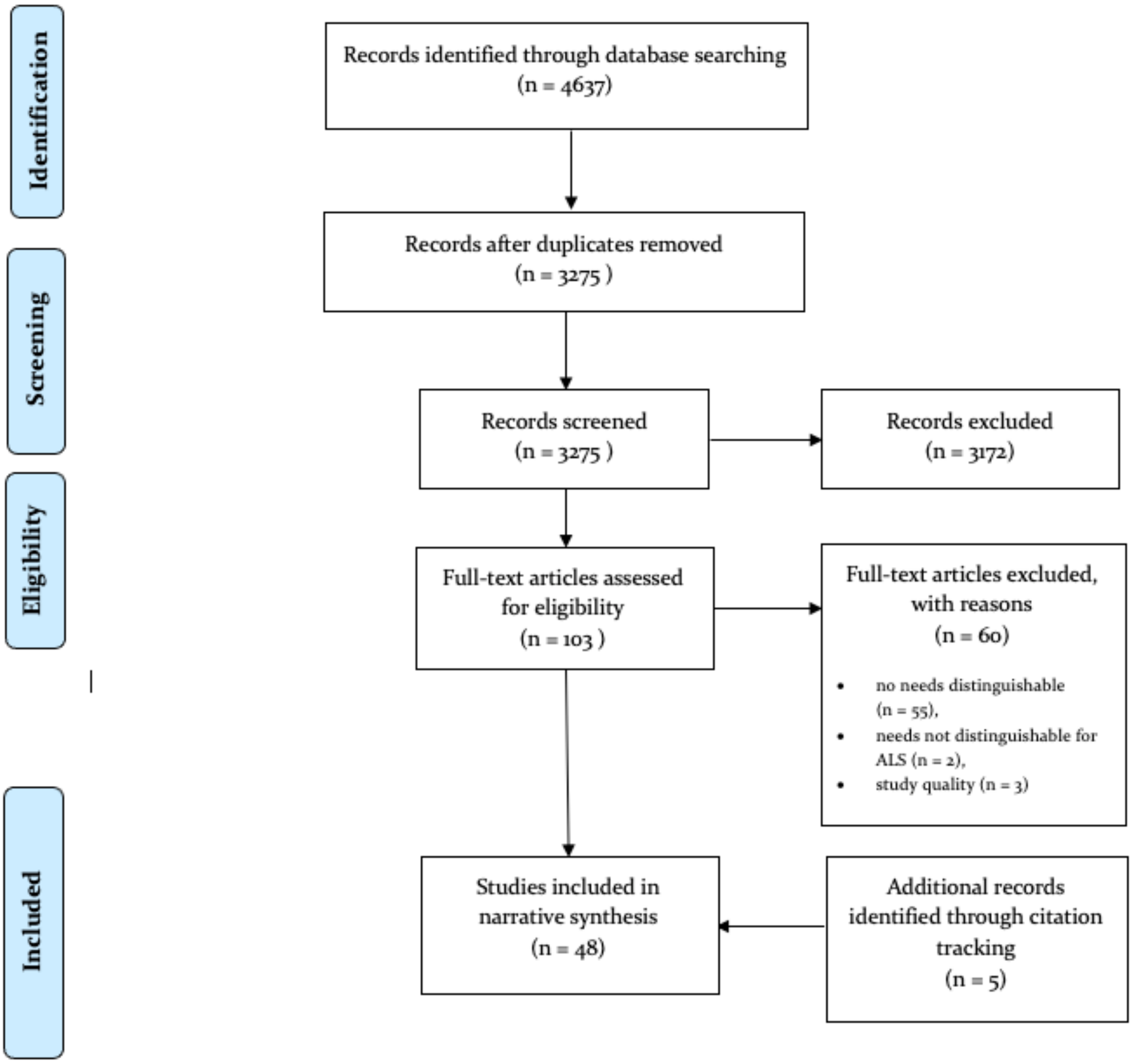
Informal caregivers provide substantial care for people with amyotrophic lateral sclerosis (pwALS), a terminal neurodegenerative disease (Mockford, Jenkinson, & Fitzpatrick, 2006; Aoun et al., 2013). Informal caregiving is 'typically provided on a voluntary basis, arising from a prior social relationship, and without financial compensation or specific training' (Hoefman, van Exel, Brouwer, 2013, p. 1106). The inexorable nature of ALS necessitates repeated adjustment of caregivers, who experience different needs (Galvin et al., 2018).

Preceding reviews on the topic neither differentiated needs by caregiving stage (Aoun, 2013; Oh & Kim, 2017; Mockford, Jenkinson, & Fitzpatrick, 2006), nor critically appraise the included studies (Mockford, Jenkinson, & Fitzpatrick, 2006; Aoun et al., 2013). They further lacked focus on pwALS' needs (Oh & Kim, 2017). This review updates the reviews with distinct stages of caregiving and with exclusive focus on informal caregivers. Williams and colleagues (2008) described four stages: 1. early coping and adjustment after diagnosis, 2. maintenance, 3. transition to the terminal stage, and 4. coping with change and loss. The last stage (4.) includes bereavement.

2. Methods

This review follows the PRISMA process for systematic reviews (Moher et al., 2010), engaging in these steps: search; screening; determining eligibility; and analysing extracted data of included studies (see Figure 1).

Figure 1: PRISMA Flow chart



Search

Our search strategy followed an adapted PICO (population, interest, comparator, outcome) scheme, which was PCO (population, context, outcome). We defined population as informal caregivers, context as ALS and outcome as needs of informal caregivers. We searched EMBASE, MEDLINE (PubMed), PsycINFO, and CINAHL as they cover relevant disciplines (biomedicine, medicine, psychology and nursing). We developed an initial list of search terms that included synonyms for caregiver (Term 1), ALS (Term 2) and needs (Term 3). A research librarian refined this search strategy (see Table 1 for all synonyms used). We adapted the search terms respectively for both searches in English and German.

Table 1: Systematic review search terms

Term 1: informal caregivers	caregiv* or carer* or spouse* or partner or husband or wife or child* or parent* or relative* or friend* or neighbo* or son or daughter or caring
Term 2: Amyotrophic lateral sclerosis	ALS or amyotrophic* or myatroph* or MND or motor neuron disease or motor neurone disease or Gehrig* or familial motor neuron or familial amyotrophic* or primary lateral sclerosis or primary lateral*

Term 3: Needs	need* or support* or unmet* or demand or wish* or require* or want* or health servic* need* or target* or goal

Screening

Two reviewers independently screened for inclusion and exclusion criteria (see Table 2). In the present review, we aimed to answer the research question 'What research evidence is there of informal caregivers' needs, that is, where do they have the capacity to benefit?'. Therefore, we excluded quality of life and burden of care studies if they did not highlight situations of need to direct future research and care practice.

Table 2: Study selection criteria

Inclusion	Exclusion
1. Needs of informal caregivers were described or were distinguishable 2. Study participants included informal caregivers of individuals with ALS or individuals with ALS, or both 3. Informal caregiving as primary unpaid caregiving for the individual	1. Needs of ALS caregivers could not be extracted separately from other groups (e.g. other neurodegenerative disorders) due to comparative designs. 2. Quality of life (QoL) and burden of care studies were excluded if

with ALS (but relied on the study author’s definitions). 4. Published in English or German 5. Published in peer-reviewed journals 6. Published from January 2000 to August 2018	they did not present specific needs of caregivers. 3. Theoretical papers that did not collect data from ALS patients and/or their caregivers 4. Papers written in languages other than English or German 5. Book chapters, dissertations, thesis, and conference abstracts 6. Peer-reviewed papers published in 1999 and earlier

Extraction of data

Two reviewers (CP and IK) independently screened for inclusion and exclusion by reading all abstracts. In case of disagreement, the reviewers discussed the abstract. Four reviewers read the resulting full texts (CP, IK, TW, LI), critically appraised the studies and in case of inclusion extracted the relevant data. Studies were appraised using either the Joanna-Briggs-Institute checklist for qualitative studies (Lockwood, Munn, & Porritt, 2015) or an adapted version of the National Heart, Lung, and Blood Institute (NHLBI) checklist for observational studies[1].

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[1] Available from https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools (10. August 2019).

Analysis

We carried out a narrative synthesis of quantitative and qualitative studies (Popay et al., 2006) because of diverse study designs. The narrative synthesis followed four steps (Popay et al., 2006 #121): '1. Developing a theoretical model of how the interventions work, why and for whom, 2. developing a preliminary synthesis, 3. exploring relationships in the data, and 4. assessing the robustness of the synthesis product' (p. 12). We did not study an intervention but hold that the method is nonetheless appropriate. As Popay et al. (2006) put it: '"narrative synthesis" […] can be used in systematic reviews focusing on a wide range of questions, not only those relating to the effectiveness of a particular intervention' (p.5).

The described needs were linked to the four stages of caregiving described previously, that is, we categorized where the described needs in the included papers belong Thereafter, we examined whether the needs described can be grouped into the Supportive Care Framework, or SCF (Fitch, 2008), which has been found to be conceptually apt in this context (Oh & Kim, 2017). The SCF consists of 'physical, emotional, social, psychological, spiritual, informational and practical changes that impact on the needs that are experienced' (Fitch, 2008, p. 8). These steps were done independently by two reviewers per paper using a data extraction form, adding to the robustness of the narrative synthesis. After these two steps, the first author examined the relationships in the data included within each SCF using descriptive inductive thematic analysis (Braun & Clarke, 2006), which helped transform the SCF groupings into themes. For the construction of themes, we assumed that a certain number of articles describing related results within each stage were necessary. If only unrelated results were available, we did not construct a theme. Additionally, the evocativeness of extracted data to relate to other extracted data was a crucial step in the formation of a theme.

3. Results

From the total of 3275 screened abstracts, 43 manuscripts met our inclusion criteria (see Figure 1). Furthermore, we tracked the citations of the 43 manuscripts in Google Scholar including five studies. Therefore, 48 studies form our study sample. Most included articles were from the UK (n=16), the EU (n=14), or from Australia (n=12). Outside of the EU, one article each from Turkey, USA, South Korea and Canada and two articles from Norway were included. In most studies, informal caregivers are family caregivers. Included studies predominantly used interview and survey methodology (see Table 3).

Table 3: Information about the included studies

Paper ID	Country	Research topic	Design	Data source	Mean quality appraisal score	Number	Caregiving stage(s)	Results related to support needs
Abdulla et al. (2014)	Germany	Information-seeking behaviour of patients with ALS and their caregivers	cross-sectional	Survey	NHLBI: 4	106 people with ALS; 100 caregivers	1,2	1. - Diagnosis came as a shock - Diagnosis communication (usefulness of diagnosis, too short information) - Need to discuss internet information 2. - Waiting time before the diagnosis was problematic
Alankaya and Karadakovan (2015)	Turkey	Home-care needs and QoL of the patients with ALS, the care burden, and QoL of caregivers, efficacy of nursing education	cross-sectional	Structured interviews/Card sorting	NHLBI: 4	30 people with ALS; 30 caregivers	2	2. - Informal caregivers have educational needs of patient care - Informal caregivers need to know how to best support the patient emotionally
Andrews et al. (2017)	Australia	Compare ALS and healthy ageing controls across a range of functional and behavioural domains using informant-	cross-sectional	Survey	NHLBI: 4	40 ALS patients and their nominated caregivers	2	2. - Poor motivation, memory dysfunction and difficulties completing activities of daily living require more carer support via

		ratings, and to examine the relationship between those difficulties and levels of caregiver burden.						direct supervision prompting or hands on care.
Aoun et al. (2012)	Australia	Experiences of MND family carers from their time as a carer of their spouse through to their bereavement	cross-sectional	Interviews	JBI: 6	16 bereaved family caregivers	1,2,3	1. - Diagnosis delivery was often burdensome 2. - The work of MND family carers is demanding - The change in relationship from spouse to caregiver 3. - Timing and access of supportive and palliative care was crucial for family carers
Aoun et al. (2017)	Australia	Experiences of family carers of people with MND in receiving the diagnosis, determine satisfaction	cross-sectional	Survey with open-ended questions	NHLBI: 4	196 family caregivers	1	1. - Important for informal caregivers was the knowledge domain of SPIKES: sharing information and empathy/warmth of the physician, who should acknowledge importance of caregiver
Baxter et al. (2013a)	UK	Attitudes, beliefs and perceptions in the first months following	cross-sectional	Interviews	JBI: 7.5	20 people with ALS; 17 family	3	3. - In person support for non-invasive

		the introduction to non-invasive ventilation				caregivers			ventilation (NIV) better than information sheet/manual - NIV caused sleep disturbance due to the noise of the machine, informal caregivers slept in a separate room
Baxter et al. (2013b)	UK	Experience of family carers and health professionals who were involved in care for ALS patients using non-invasive ventilation	cross-sectional	Interviews	JBI: 7	9 family caregivers	3	3.	- Informal caregivers have misunderstandings concerning NIV functioning (unexpected speed of deterioration, caregivers did not know signs and caregivers did not understand that NIV does not prevent respiratory failure) - Emergency admission was burdensome for informal caregivers (knowing when to call an ambulance, attempts to resuscitate were distressing) - Turning off the machine after death was distressful
Bentley and O'Connor (2016)	Australia	Perceptions of family carers of people with MND	cross-sectional	Interviews	JBI: 7.5	12 family caregivers	3	3.	- Accessing appropriate support

									- Accessing information - Feeling prepared for the death
Brown et al. (2005)	UK	Investigate whether services met the needs of patients with MND and their carers, to explore preferences for service delivery and compare these with services provided locally	cross-sectional	Interviews	JBI: 5	11 people with ALS; 9 family caregivers	2	2.	- Increased knowledge of MND among health & social care professionals - Better coordination of services - Faster response - Availability of respite and hospice care - Chance to give feedback
Bruletti et al. (2015)	Italy	Strain and needs in caregivers of advanced ALS patients and correlate burden with patients clinical condition and caregivers sociodemographic status	longitudinal	Survey	NHLBI: 4.5	T0: 58 family caregivers; T1: 39 family caregivers; T2: 13 family caregivers	3	3.	- High level of needs on the Caregiver Needs Assessment - The authors conclude that caregivers need more information about the disease
Chiò et al. (2008)	Italy	Information preference and satisfaction with diagnosis communication as well as related information seeking behaviour in ALS patients and caregivers	cross-sectional	Structured questionnaire	NHLBI: 4.5	60 family caregivers	1	1.	- Informal caregivers may experience discouragement after diagnosis but appear encouraged during the appointment where the diagnosis was given - Caregiver have higher interest to receive information about variability

								and prognosis - High interest in research on therapies - Patients and caregivers searched for more information elsewhere
Clabburn and O'Brien (2015)	UK	Explore how providing care for a parent with MND impacts upon a young person's life	cross-sectional	Interviews	JBI: 7	7 young (related) caregivers	1,2,4	1. - After being diagnosed with MND, parents withheld information 2. - Changing family roles - Implications upon education - Implications upon social life 4. - Implications upon education (lack of learning partner)
Dawson and Kristjanson (2003)	Australia	Identify the particular needs of families of people with MND towards the end of their lives	cross-sectional	Interviews	JBI: 7.5	5 family caregivers	3	3. - Need for support with reactions and responses at the end of life: Grief everyday, loss of social networks, of retirement years and dreams, fearing each crisis could mean the end, watching life in reverse, living with limits, and worry about the future. - Need for support in relation to

									the health system as crossing points and getting lost in the system - Getting help in general - Need for access to palliative care - Informational need to understand the condition
Foley et al. (2016)	Ireland	Patients' experiences of informal carer burden	cross-sectional	Interviews	JBI: 6.5	34 people with ALS	2	2.	- Mediating needs: Patients decide their care to take pressure off caregivers/decide in agreement with family caregivers' preference of care in order to help them adjust. - Patients also render support to their family mainly emotional support
Galvin et al. (2016)	Ireland	Multidimensional nature of caregiver burden in ALS through the use of quantitative and qualitative data, increasing the type and range of information available for analysis, for complimentary and additional coverage.	cross-sectional	Survey/Interviews	JBI: 5.5	81 family caregivers	2	2.	- Emotional needs relating to fear of the future, the feeling of dependence, and psychosocial and emotional well-being, and various other emotions - Impact on social life, relationships with others, time restriction (role, personal

									strain & guilt) - Help with management and assistance with competing responsibilities and practicalities of the condition - Lack of recognition
Galvin et al. (2017)	Ireland	The journey from the first problem symptoms to diagnosis from the perspective of informal caregivers providing care to people with ALS	cross-sectional	Interviews	JBI: 8.5	74 family caregivers	2	2.	- Problems and symptoms noticing and reaction and response before diagnosis - Uncertainty, worry, search for information, connect with healthcare professionals - Interaction with healthcare services, convince patient to seek help, referral routes, tests/interventions, misdiagnosis, not being listened to
Galvin et al. (2018)	Ireland	Needs of informal ALS caregivers across the caregiving course	longitudinal	Interviews	JBI : 8	T0:81 family caregivers; T1:56 family caregivers; T2:41 family caregivers	2	2.	- Need for external support and assistance - Support with psychological-emotional factors - Need for the patient to accept services - Need for nothing or a cure

Grimby, Johansson, and Johansson (2015)	Sweden	Describe the outcome of Anticipatory Grief Scale measurement among close relatives to persons suffering from ALS and MS	Cross-sectional	Survey	NHLBI : 4	89 ALS and MS primary caregivers	2	2.	- 39% of ALS primary caregivers need to talk to somebody outside the family and hospital staff
Hecht et al. (2003)	Germany	The total burden, the components of burden and the relationships between subjective burden of care and functional impairment of the patient	cross-sectional	Structured interviews	NHLBI: 4	37 family caregivers; 37 people with ALS	2	2.	- Informal caregivers need help with personal and social restrictions components on the Cost of Care Index (CCI)
Herz et al. (2013)	Australia	Experiences and perceptions of carers and former carers of people with MND with emphasis on the later stages of the disease	cross-sectional	Focus Group Discussion	JBI: 7	8 former family caregivers; 3 current family caregivers	3,4	3. 4.	- Need for an active role of the general practitioner and MND associations - Access help and palliative care especially to help with unremitting care - Need for financial help due to financial burden - Emotional costs to the carer and needs relating to: Love, 'Trapped and drowning', suspension of own needs

								- Support with 'return to the living'
Hill and Wilson (2013)	UK	Experiences of people with MND and their informal carers who had accessed crisis intervention services during a medical emergency	cross-sectional	Interviews	JBI: 7	7 family caregivers; 7 people with ALS	3	3. - Reluctance to call for assistance, maintaining independence - Impact of crisis events - Lack of coordination and responsiveness of services - Lack of awareness of MND/spouses needs - Lack of information about services
Kristjanson et al. (2006)	Australia	Needs for supportive care/palliative care services of people in Australia with MND, MS, HD, PP and the needs of their carers	cross-sectional	Survey	NHLBI: 4	105 family caregivers; 119 people with ALS	3	3. - Need for accessible services - Need for equipment for daily living - Information about how to provide care - Reliable, ongoing dependable support workers - Financial assistance for care
Larsson et al. (2015)	Sweden	Relatives' experiences of patient care and the support they themselves received during the course of the disease	cross-sectional	Interviews	JBI: 7.5	15 family caregivers	2,4	2. - 'Patient-centered care inspired a feeling of security' - Informal caregivers need good treatment provided by the staff, help and support from the staff, knowledge

									of ALS among the staff, availability of the staff/care, continuity of care. - ‘Support was available, but different factors influenced its use by the relatives’: availability, different support needed/demanded (talk to other relatives, need for information and knowledge about the disease and care - Healthcare services need to take an active role in providing support.
								4.	- ‘a need to talk after the patient had died’
Lerum et al. (2016)	Norway	Family caregivers' work and sense of responsibility, exploring family caregiver's accounts of caring for a family member with MND	cross-sectional	Interviews	JBI: 8	25 caregivers	2	2.	- Five lines of work for which informal caregivers need support: immediate care work, seeking information and clarity, managing competing obligations, maintaining normality, and managing external

									resources and assistance
Locock and Brown (2010)	United Kingdom	Attitudes to peer support among MND patients and family caregivers	cross-sectional	Interviews	JBI: 6.5	22 caregivers; 48 people with ALS	2	2.	- Getting advice from peers as self-help, camaraderie, and becoming part of special group was appreciated.
Martin and Turnbull (2001)	Canada	Impact of ALS on surviving family members, to assess their needs for ongoing support in the clinic and other agencies and their ongoing involvement in ALS-related issues	cross-sectional	Survey	NHLBI: 4.5	27 caregivers	2,4	2. 4.	- Need for increased support in home care - Respite care and financial support - Experienced self-defined burn-out - Ongoing questions about the disease and wishes to receive literature/information
Murray et al. (2016)	Australia	Caregiver perspectives on the acceptability and impact of advance care planning, documented in a letter format	cross-sectional	Semi-structured interviews	JBI: 6.5	10 caregivers; 8 people with ALS	3	3.	- Appropriate timing to commence advance care planning - Readiness of patients and caregivers for uptake of advance care planning
Neudert et al. (2001)	Germany/United Kingdom	Course of the terminal phase in ALS	cross-sectional	Structured interviews/Medical records	NHLBI: 4.5	121 German caregivers; 50 UK caregivers (2 professional carers)	3	3.	- Need for an early discussion of advance directives with ALS patients and their caregivers (often

									resuscitation attempts).
Ng et al. (2011)	Australia	Disability profile and healthcare needs for persons with MND (pwMND) in an Australian sample from the perspective of the patients and caregivers to identify current gaps in the knowledge and service provision	cross-sectional	Structured interviews/Medical records	NHLBI: 5	37 caregivers; 44 people with ALS	2	2.	- Formal paid care for personal assistance/carer support housework - Respite care - Emotional support - Preparation for special diet
O'Brien et al. (2011a)	UK	Personal perspectives of the diagnostic experience for people with ALS/MND and their family carers identifying issues that could impact positively or negatively on these experiences	cross-sectional	Interviews	JBI: 7	24 people with MND; 18 current family carers; 10 former family carers	1, 2	1. 2.	- Need for a good communication of diagnosis: negative and positive experiences of receiving the diagnosis - Responding to the diagnosis: informal caregivers need time to cope, prepare to face diagnosis, relief having a label, emotional reactions, information needs, communicating the diagnosis to the family. - Lack of recognizing ALS diagnosis (by oneself and by the physician

								physician), diagnosis delays, non-neurological referrals - Primary care misses symptoms, symptoms hidden by other conditions
O'Brien et al. (2011b)	UK	Views of people with MND and family carers regarding multidisciplinary team (MDT) working	cross-sectional	Interviews	JBI: 7	24 people with MND; 18 current family carers; 10 former family carers	2	2. - Having one point of access: joint consultations between health professionals would be an improvement - Difficulties with accessibilities and having to travel long distances to the MND clinic - Lack of clarity regarding the roles and responsibilities of health and social care professionals involved in patient care which could be confusing for people unfamiliar with the system, - Regular follow-up: continuity of care and of social workers, regular contact to GP.
O'Brien et al. (2012)	UK	Carers' need for, and use of, support services are not fully	cross-sectional	Interviews	JBI: 8	18 current & 10 former caregivers	2,4	2. - Information needs regarding impact, progression

		understood; this study aimed to explore, from a qualitative perspective, the views of current and former family carers of people with MND						and time to look for information - Access to support services, lack of signposting - Training to become a more competent carer - Knowledge of roles and responsibilities of professionals - Financial help due to loss of finances - Lack of continuity of healthcare service provision - Respite care needed, but is an invasion of lives, need to repeatedly instruct the respite carers, reliability of respite care. Accessibility of respite, guilt about leaving, patients sometimes unwilling to participate - Need for counselling 4. - Counselling after bereavement
O'Brien and Preston (2015)	UK	Experiences of hospitalisation that occurred after a diagnosis of MND was made (i.e., not related to the	cross-sectional	Interviews	JBI: 7.5	3 current; 18 former caregivers	2, 4	2. - Lack of knowledge among staff members; informal caregivers need to stay and educate - Lack of basic care

		diagnostic process) from the perspective of family carers of people diagnosed with the illness.						4.	and communication, caregivers still perform personal care (hygiene) - Admission as personal failure/reluctance to admission. - Negative hospital experience impacts last memories
O'Connor et al. (2018)	Australia	The aim of the study was to describe the experiences of family carers of people with MND in receiving the diagnosis in order to inform and improve ways in which the diagnosis is communicated.	cross-sectional	Survey open question	JBI: 7.5	190 current caregivers	1	1.	- Frustration with the diagnosis (misdiagnosis, shock of diagnosis) - Giving information (pacing and planning) - Need for a caring physician - Private setting and time for communication of diagnosis
Oh and Schepp (2013)	South Korea	The purpose of this ethnographic study was to describe the lived experience of wives in South Korea whose husbands were diagnosed with amyotrophic lateral sclerosis.	cross-sectional	Interviews	JBI: 7	11 wives, one mother and 2 male caregivers	2	2.	- New roles as the head of family and guardian - Sexual relations - Relationship with in-laws - Becoming family decision-maker
Olsson Ozanne et al.,	Sweden	Illuminate factors that facilitate and hinder	cross-sectional	Interviews	JBI: 7	13 caregivers; 14 people	2	2.	- Relating to "self": need for

2012 et al. (2012)		the manageability of living with ALS in patients and next-of-kin				with ALS		acceptance and being in the present to cope with fear, hate and one's own ill health - Relating to "family": need for understanding and support - Communication needs - Managing children/practical liabilities - Need "Real presence" not absence of support
Oyebode et al. (2013)	UK	Experience of living with, and caring for, a partner with MND.	cross-sectional	Interviews	JBI: 7	8 partner caregivers	2	2. - Need for respite to keep up with social life/time for self - Need for sensitive delivery of information - Use of percutaneous endoscopic gastrostomy (in public restaurants)
Ozanne et al. (2015)	Sweden	Illuminate experiences of finding meaning in life among spouses of people with ALS	cross-sectional	Interviews	JBI: 7	13 caregivers; 14 people with ALS	2.	2. - Living an imprisoned life, feeling lonely, considering life as unfair and incomplete, mourning a lost future
Ozanne and Graneheim (2018)	Sweden	Illuminate patients and spouses experiences if comprehensibility in ALS from a long-term perspective, when symptoms	cross-sectional	Interviews	JBI: 7.5	13 caregivers; 14 people with ALS	1,2	1. - Losing one's foothold, informal caregivers need information. 2. - Feeling uncertainty

		appeared before diagnosis and when the diagnosis was						- Information about diagnosis - Emotional needs: caregivers lived in fear - Unclear pathogenesis: ‘looking for reasons why’
Peters et al. (2012)	UK	Relationship between support provided by health and social care services and MND caregivers and patient well-being	cross-sectional	Survey	NHLBI: 4	407 caregivers	2	2. - Caregivers' experiences not being valued by health and social care services - Lack of involvement in care planning
Ray and Street (2006)	Australia	Caregiver experiences of bodywork and the associated emotional labor	cross-sectional	Interviews and participant observation	JBI: 6	18 caregivers	2	2. - Caregivers are emotionally vulnerable and need emotional support, but have difficulty asking for it (loss of face).
Ray, Brown, and Street (2014)	UK/Australia	To examine the ways, family caregivers of people living with motor neurone disease (MND) experienced the dying of their relative	longitudinal	Secondary analysis of interview data	JBI: 9	29 family caregivers	3,4	3. - Discussing and planning end-of-life care, preparing for death - Constructing death as the final part, taking time to say goodbye, calmness and peace 4. - Shock from unexpected dying - Unprepared for rapid deterioration - Difficulty reorganizing social

									world post-caregiving
Sandstedt et al. (2018)	Sweden	Caregiver experience, HRQL, and life satisfaction in informal caregivers to patients with ALS	cross-sectional	Survey	NHLBI: 4	49 informal caregivers	2	2.	- Relevant dimensions on the Caregiver Reaction Assessment: Own ill health 'Health problems', 'disrupted schedule', 'caregiving esteem'
Siewers et al. (2013)	Norway	ALS patients', their family carers', and health professionals' experiences with using a mechanical in-exsufflation device in the home setting	cross-sectional	Interviews	JBI: 7.5	3 informal caregivers	3	3.	- Training extending the plain instruction on how to handle the machine, so that those who were involved had the qualifications to do adjustments if necessary - Ability to use the device competently
van Teijlingen et al. (2001)	UK	Current use of services and the perceived needs of the MND population in Scotland.	cross-sectional	Survey	NHLBI: 4.5	153 informal caregivers	2	2.	- Need for help with housework - General help - More freedom to go out - Knowing some is available if needed
Weisser et al. (2015)	UK	Experiences of family caregivers of people with MND/ALS	longitudinal	Interviews	JBI: 7	10 informal caregivers	1,2	1.	- Particularly at the beginning of the illness, psychological needs; need for connectedness and

								communication 2. - Need to actively seek help - Access to agencies - Timely information
Whitehead et al. (2012)	UK	Experiences of people with MND, current and bereaved carers in the final stages of the disease and bereavement period	cross-sectional	Interviews	JBI: 7.5	18 current caregivers; 10 bereaved caregivers	3, 4	3. - Need for advance care planning (i.e. Preferred priorities of care) - Need for good quality of services - Need for increased and coordinated palliative care - Discussion with professionals appreciated 4. - Bereavement support
Williams et al. (2008)	USA	Needs of ALS family caregivers	cross-sectional	Interviews and survey	JBI: 6.5	19 caregivers	2	2. - 109 statements of needs in the form of “I need …”. Needs were not synthesized.

All included studies mentioned numerous needs which are synthesized and grouped into four caregiving stages (Williams et al., 2008). Most studies were not directed towards a specific stage and best fitted the maintenance category, which became an overall category. We also point to challenges before diagnosis there. An overview of themes is presented in Table 4. The order of presentation is not hierarchical.

Table 4: Overview of themes

Stages*	Themes
1.Early coping and adjustment after diagnosis	- Process of diagnosis - Information about diagnosis - Response to diagnosis
2. Across the caregiving course (maintenance)	- Informational needs - Emotional support - Formal support with care at home - Needs relating to healthcare services - Financial support - Role, social, and other needs - Dietary needs
3. Transition to the terminal stage	- Information provision and access of services - Contact with healthcare practitioners - Emotional impact of dying on the informal caregiver - Assistive technology at the end of life - Advance planning
4. Coping with change and loss after bereavement	*Due to lack of studies no themes could be generated*

*Adapted from Williams, M.T., Donnelly J.P., Holmlund T., & Battaglia, M. (2008) ALS: Family caregiver needs and quality of life. *Amyotroph Lateral Scler*. 279-286, 9(5).

Early coping and adjustment after diagnosis (stage 1)

The diagnosis of ALS comes as a shock to informal caregivers (O'Brien et al., 2011; Abdulla et al., 2014; O'Connor, Aoun, & Breen, 2018; Ozanne & Graneheim, 2018). This stage centers around diagnosis of ALS as a life-threatening illness: getting the diagnosis, understanding it and responding to it.

Process of diagnosis

The needs of informal caregivers primarily relate to the communication of diagnosis (Abdulla et al., 2014), which was sometimes experienced as negative (O’Brien et al., 2011) and other times as a relief of having a label (O'Brien et al., 2011; Aoun et al., 2012). It was noted that informal caregivers need a ‘caring physician’ (O'Connor, 2008). Similarly, sharing of information, physician empathy and acknowledging the importance of the informal caregiver at diagnosis are important (Aoun et al., 2017). An appropriate private setting for the communication of the diagnosis (O'Connor, Aoun, & Breen, 2018) and emotional preparation are necessary to receive the diagnosis (O'Brien et al., 2011).

Information about diagnosis

Comprehensive information about ALS is needed to understand its implications (O’Brien et al., 2011; Abdulla et al., 2014; Ozanne & Graneheim, 2018). Physicians should pace and plan information provision (O'Connor, 2018). Informal caregivers look for information from other sources (Chiò et al., 2008), which they wish to discuss (Abdulla et al., 2014). Underlined in one study was the specific desire of informal caregivers for information about disease variability and prognosis, and research on therapies (Chiò et al., 2008).

Response to diagnosis

Informal caregivers experienced the diagnosis appointment as encouraging, but experienced distress after it ended (Chiò et al., 2008). In response, they need time to cope (O'Brien et al., 2011) and to communicate the diagnosis to the family (O'Brien et al., 2011). Weisser et al. (2015) found that at the beginning of the illness, psychological needs for connectedness and communication were universal themes.

Across the caregiving course (stage 2)

While supportive needs for informal caregivers can only be described after a diagnosis of ALS has been established, the time to diagnosis presents the first challenge for family and next-of-kin. From the beginning of symptoms, they notice problems and symptoms, and experience uncertainty and worry (Galvin et al., 2017; Ozanne & Graneheim, 2018). They often contact healthcare professionals (HCPs) and urge pwALS to seek help (Galvin et al., 2017). Missed symptoms by general practitioners (GPs), unnecessary tests or interventions, and misdiagnosis are problematic for informal caregivers (O'Brien et al., 2011). Minimization of time to specialist referral and diagnosis were deemed crucial (O'Brien et al., 2011; Abdulla et al., 2014).

After diagnosis and across the caregiving course, Williams et al. (2008) ascertain that 'family caregivers focus on establishing as much control over day-to-day life as possible' (Williams et al., 2008).

Information needs

Seeking information and clarity is one ‘line of work’ (Lerum, Solbraekke, Frich, 2016) of informal caregivers. Information about providing care is urgently needed (Martin & Turnbull, 2001; Kristjanson, Aoun, & Yates, 2006; Alankaya, 2015; Larsson et al., 2015). Informational needs surrounding impact and progression of ALS exist (Kristjanson, Aoun, & Yates, 2006). Informal caregivers wish for a sensitive delivery of (Oyebode, Smith, & Morrison, 2013) and for adequate time to understand information (O’Brien et al., 2012). Sometimes, informal caregivers lack guidance to navigate the healthcare system and knowledge about roles and responsibilities of HCPs, especially if they are unfamiliar with the healthcare system (O’Brien et al., 2011; O’Brien et al., 2012). One study (Alankaya, 2015) showed an unmet need for information about providing emotional support. Informational needs might be best resolved by talking to peers (Locock, 2010; Larsson et al., 2015).

Emotional support

Emotional support is a major theme in this stage (Ng, Talman, & Khan, 2011; Galvin et al., 2016; Galvin et al., 2018). Caregivers are emotionally vulnerable and need support but have difficulties asking for it due to loss of ‘face’ (Ray & Street, 2006). A major concern is the fear of the future and various emotions experienced during caregiving (Ozanne, 2015; Galvin et al., 2016; Ozanne & Graneheim, 2018). Lack of recognition and the feeling of not being valued likewise impact informal caregivers’ emotional well-being (Peters et al., 2012; Galvin et al., 2016), and might run down caregiving esteem (Sandstedt, 2018).

Formal support with care at home

Several studies found a need for increased support for home care (Martin & Turnbull, 2001; Ng, Talman, & Khan, 2011; Aoun et al., 2012; Galvin et al., 2018; van Teijlingen, Friend, & Kamal, 2001) and general help (van Teijlingen, Friend, & Kamal, 2001), immediate care work and managing external resources (Lerum, Solbraekke, Frich, 2016), formal paid care for personal assistance (Ng et al., 2011; Galvin et al., 2018), and assistance with management of practicalities (Galvin et al., 2016).

Several studies revealed that informal caregivers have a need for respite care (Martin & Turnbull, 2001; van Teijlingen, Friend, & Kamal, 2001; Ng et al., 2011; O'Brien et al., 2012), which is defined as a short period of time 'where the cared-for person was admitted […] to a care facility such as a hospice' (O'Brien et al., 2012) or as paid caregivers providing care to pwALS (Ng, Talman, & Khan, 2011).

Respite helps to 'keep up with social life' (Oyebode, Smith, & Morrison, 2013) and to have time for oneself (van Teijlingen, Friend, & Kamal, 2001; Oyebode, Smith, & Morrison, 2013), which helps to cope with a 'disrupted schedule' (Sandstedt, 2018). It was reported that respite care must be reliable and accessible (O'Brien et al., 2012). In contrast, respite care can be an invasion of the private life and can be burdensome if respite staff needs instruction (O'Brien et al., 2012).

Needs relating to healthcare services

Informal caregivers experience different needs for healthcare services (Dawson & Kristjanson, 2003; O'Brien et al., 2011). Generally, accessible healthcare services are needed from an informal caregivers' perspective to provide equipment for daily living and reliable support workers (Kristjanson, Aoun, & Yates, 2006).

Central to the contact with healthcare service is care coordination. Informal caregivers appreciate better coordination of healthcare services (Brown, Lattimer, & Tudball, 2005), joint consultations between HCPs, and having a centralized point of access to these services (O'Brien et al., 2011). Additionally, continuity of care, regular follow-up with healthcare and social workers was as important as regular contact to the local GP (O'Brien et al., 2011; O'Brien et al., 2012; Larsson et al., 2015).

Accessibility and close proximity of ALS clinics are needed to avoid travelling long distances (O'Brien et al., 2011). If pwALS were at the centre of care, informal caregivers felt secure, which can be understood as a psychological and emotional need of informal caregivers (Larsson et al., 2015). Likewise, informal caregivers need specialized treatment for pwALS provided by experienced HCPs, and their explicit support in providing informal care (Larsson et al., 2015). Informal caregivers want their HCPs and other support workers to be well informed about ALS (Brown, Lattimer, & Tudball, 2005; Larsson et al., 2015). In case of hospital admission, informal caregivers perceived a lack of knowledge among hospital staff members and hence continued to perform personal care and train hospital staff members (O'Brien & Preston, 2015). Informal caregivers need to be supported by HCPs pro-actively (Larsson et al., 2015) but at the same time support offered to informal caregivers might not be the support they really need (Larsson et al., 2015).

Financial support

Financial support was discussed in three studies, however, in those studies it was not the main aim or finding. Thus, it was presented that informal caregivers mention financial needs due to loss of income (O'Brien et al., 2012), and hence require financial assistance for care (Martin & Turnbull, 2001; Kristjanson, Aoun, & Yates, 2006).

Dietary needs

Special needs of informal caregivers also relate to the dietary needs of the pwALS. In one study, the ability to use percutaneous endoscopic gastrostomy (PEG) feeding in public restaurants was appreciated by informal caregivers (Oyebode, Smith, & Morrison, 2013). Additionally, informal caregivers expressed the need for support regarding the preparation of special diets (Ng, Talman, & Khan, 2011).

Role, social, and other needs

Some needs of informal caregivers relate to their person and their role as caregivers (Galvin et al., 2016; Aoun et al., 2012). Most studies use the umbrella term informal caregivers, meaning mostly family, often spousal, caregivers. With the caregiving role might also come the role as the new head of family, guardian and decision maker (Oh & Schepp, 2013). The situation of young carers providing care to a parent highlights distinct needs, as Clabburn and O'Brien (2015) show: changing family roles, implications upon education and upon social life, and parents withholding information, among others.

Adult informal caregivers manage competing obligations, for example to children (Olsson Ozanne et al., 2012; Galvin et al., 2016), and experience personal strain additionally to role strain (Galvin et al., 2016), while ‘maintaining normality’ (Lerum, Solbraekke, & Frich, 2016). Part of maintaining normality is keeping up social relationships (Galvin et al., 2016), which are restricted when providing care (Hecht et al., 2003; Galvin et al., 2016). Informal caregivers experience a need to talk and for camaraderie in self-help groups as a form of peer-support (Locock, 2010; Grimby, Johansson, & Johansson, 2015). Further needs arise when informal caregivers themselves become ill (Olsson Ozanne et al., 2012, 2012; Sandstedt, 2018). If there is a pre-existing sexual relationship with the pwALS, maintaining it can become a burden (Oh & Schepp, 2013).

Factors related to the pwALS, for example the acceptance of services (e.g. respite care), help informal caregivers to adjust (Galvin et al., 2018; O’Brien et al., 2012). Informal caregivers benefit from understanding, support and communication within the family (Olsson Ozanne et al., 2012, 2012) but can also be bothered by relationships with in-laws, one study from Korea found (Oh & Schepp, 2013). Communication within the family permits to mediate needs in the caregiving relationship as some pwALS decide to change the provision of care services to take pressure off caregivers (Foley, Timonen, & Hardiman, 2016).

If pwALS undergo behaviour change and neurocognitive impairments, these can be burdensome for informal caregivers. Specifically, Andrews and colleagues (Andrews et al., 2017) found that dysfunction with everyday skills, motivation and memory ‘requires caregivers to provide more direct supervision, prompting, or hands on assistance day-to-day’.

On the positive side, sometimes pwALS function as caregivers and provide emotional support to their family (Foley, Timonen, & Hardiman, 2016). Galvin and colleagues (Galvin et al., 2018) found that some informal caregivers have a need for a cure or for ‘nothing’. The latter could mean either satisfaction with support or the difficulty to receive help during caregiving for an intractable terminal disease.

Transition to the terminal stage (stage 3)

Loss and dying dominate this stage, and informal caregivers are actively working against it. Accordingly, Bruletti et al. (2015) found that during the transition to the terminal stage most informal caregivers continue to have high level of needs, but that informational and emotional needs are less important then.

Information provision and access to services

Nonetheless, informal caregivers still need access to information about ALS (Bruletti et al., 2015; Bentley & O’Connor, 2016) in its last stage. At that time, informal caregivers wish for access, the increased provision, and better coordination of palliative care services (Dawson & Kristjanson, 2003; Brown, Lattimer, & Tudball, 2005;Whitehead et al., 2012; Bentley & O’Connor, 2016). Palliative care services are ‘the active total care offered to a person with a progressive illness and his/her family, concentrating on the quality of life and the alleviation of distressing symptoms’ (Dawson & Kristjanson, 2003). Timing of palliative care is crucial for informal caregivers. Generally, informal caregivers lack information about the availability of services and want services to be responsive to their needs (Hill & Wilson, 2013).

Contact with healthcare practitioners

At the end of the pwALS' life, the chance to give feedback and the opportunity to discuss with HCP is important (Brown, Lattimer, & Tudball, 2005; Whitehead et al., 2012; Bentley & O'Connor, 2016). Informal caregivers need healthcare and social care practitioners to have appropriate knowledge about ALS, for example at hospital admission to non-specialist wards (Brown, Lattimer, & Tudball, 2005; O'Brien & Preston, 2015). GPs and ALS associations play a crucial role in helping informal caregivers in the palliative phase (Herz, McKinnon, & Wilson, 2013). Some informal caregivers viewed hospital admission of the pwALS as a personal failure, and were thus hesitant to admit them (O'Brien & Preston, 2015). Hill and Wilson (Hill & Wilson, 2013) revealed that informal caregivers are reluctant to call for assistance because they wanted to remain independent in the care for the pwALS. Similarly, in emergencies informal caregivers need to know when to call an ambulance. Attempts of cardiopulmonary resuscitation were distressing for informal caregivers (Baxter et al., 2013).

Emotional impact of dying on informal caregivers

Not only crisis events (Dawson & Kristjanson, 2003; Hill & Wilson, 2013), but also the dying of the pwALS have an emotional impact on the informal caregiver. Informal care in the last stages of ALS is 'unremitting care' and as everyday grief (Dawson & Kristjanson, 2003; Herz, McKinnon, & Wilson, 2013; Grimby, Johansson, & Johansson, 2015), which makes respite and hospice care crucial (Brown, Lattimer, & Tudball, 2005).

There are emotional costs in caring for a pwALS in the last days: informal caregivers feel 'trapped and drowning' (Herz, McKinnon, & Wilson, 2013) while 'watching life in reverse' (Dawson & Kristjanson, 2003), have problems accessing help or to feel loved, and suspend their own needs (Herz, McKinnon, & Wilson, 2013). Losses of social networks, retirement years, and dreams affect informal caregivers (Dawson & Kristjanson, 2003). Feeling prepared for the death of the pwALS is both a practical, e.g. being able to care for a dying person at home, and informational needs, e.g. knowing about prognosis and signs of deteriorations (Bentley & O'Connor, 2016).

Assistive technology at the end of life

If the pwALS uses non-invasive ventilation (NIV), informal caregivers need education and training about its functioning (Baxter et al., 2013). Baxter and colleagues (2013) reported that after pwALS died with a mask, turning off the machine in the home setting was distressing for informal caregivers.

For the education about NIV, in-person support (e.g. through a HCP) is better than textual information (Baxter et al., 2013). Informal caregivers were burdened by the noise of the NIV machine, which led to sleep disturbance and forced them to sleep in a separate room (Baxter et al., 2013). For the use of an In-Exsufflator, informal caregivers require extended training to perform necessary adjustments and use the device competently (Siewers, Holmøy, & Frich, 2013).

Advance planning

While advance planning is crucial for emergencies and end-of-life circumstances, it should be prepared well before the last stages of ALS. Appropriate timing and readiness to commence advance care planning are important (Ray, Brown, & Street, 2014; Murray et al., 2016). Informal caregivers need early discussions of advance care planning with pwALS, as there are often unwanted resuscitation attempts (Neudert et al., 2001), and they want to be involved in this process (Peters et al., 2012; Whitehead et al., 2012).

Coping with change and loss after bereavement (stage 4)

Only three studies described needs of informal caregivers of deceased pwALS. After death of pwALS, informal caregivers had problems to 'return to the living' (Herz, McKinnon, & Wilson, 2013). Whitehead et al. (2012) reported that bereavement support was an unmet need. Access to counselling services after bereavement is crucial (Whitehead et al., 2012), as there is 'a need to talk after the patient had died' (Larsson et al., 2015). In case of death in a hospital, (negative) hospital experiences might affect lasting memories (O'Brien & Preston, 2015).

4. Discussion

This systematic review synthesizes needs across four caregiving stages. In the first stage, needs of informal caregiver centre on the diagnosis. The second stage comprises diverse themes of support across the caregiving course (formerly maintenance stage). Preparation for the death of the pwALS weaves through all themes in stage three. Finally, stage four did not include enough results to satisfy the construction of themes. However, certain needs of informal caregivers are noted relating to the difficult return to life after death of the pwALS. The limited information that we found for the bereavement stage points to the lack of available knowledge regarding informal caregivers' differentiated needs once they no longer are an informal caregiver.

In the distribution of studies across caregiving stages, bereavement is addressed less often than the other caregiving stages, possibly, as these needs are outside of healthcare contact for pwALS. Most studies describe needs that are relevant across the caregiving course. These diverse needs might best be met by HCPs specialising in ALS care. Nonetheless, this review did not evaluate access to support, which might be very different.

Limitations

Our review is limited to the search terms and databases used, thus, relevant studies may have missed our attention. We tried to ascertain comprehensiveness by further screening studies citing the included manuscripts.

Furthermore, issues known from clinical work, e.g. the role of children or medical assistance in dying (Gamondi et al., 2015; Gamondi et al., 2018), have marginally been raised and might not be prevalent in the research literature or might not be associated with supportive needs. We also found little evidence for financial and peer support needs, which could be an artefact of the included studies. Indeed, literature published after our inclusion date refers to peer support (de Wit et al., 2019). Additionally, missing in the screened literature are the needs of informal caregivers who are confronted with a pwALS' wish to die e.g. by assisted suicide, euthanasia, or suicide.

Finally, our narrative synthesis uses descriptive inductive thematic analysis, lacking a) a more theoretical understanding of evolving needs across caregiving stages and b) higher levels of evidence.

Future research

Aoun et al. (2013) stipulated that ALS informal caregiving research should move from descriptive to interventional research. Most caregiving needs presented in this review are out of the scope of psychological interventions, e.g., financial support, support with formal care at home, and diagnosis-related needs. Thus, research is needed on psychosocial and service-related interventions and their timing, pacing, and planning to address the caregiving needs. This systematic review underscores that needs of informal caregivers differ across the caregiving stages and this differentiation should direct future research and care practice. The first and last stages of caregiving remain understudied, and studies are lacking from non-OECD countries. Table 5 contains a comprehensive checklist for future research.

Table 5: Checklist for future informal caregiving studies

1.	Providing comprehensive accounts of informal caregivers needs
2.	Differentiating symptom onset and ALS subtype
3.	Differentiating neurocognitive and behavioral impairments
4.	Differentiating personal characteristics of people with ALS and informal caregivers
5.	Differentiating stage of caregiving, preferably in a longitudinal design
6.	Extending the scope of interventions beyond psychological interventions

5. Conclusion

Diverse needs of informal caregivers ask for flexible modes of support. Healthcare professionals should pay attention to the different caregiving stages to respond to those differentiated needs. Further research is necessary for the bereavement phase and in non-OECD countries.

Figures and tables

Figure 1: PRISMA Flow chart

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Chapter 4.

Supportive needs of informal caregivers of people with amyotrophic lateral sclerosis in Switzerland: A qualitative study

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Supportive needs of informal caregivers of people with amyotrophic lateral sclerosis in Switzerland: A qualitative study

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Supportive needs of informal caregivers of people with amyotrophic lateral sclerosis in Switzerland: A qualitative study

Abstract

Objective: This study explores the supportive needs of informal caregivers of people with amyotrophic lateral sclerosis (ALS) in Switzerland.

Method: We conducted semi-structured interviews with 9 caregivers currently providing care to a person with ALS, 14 bereaved informal caregivers, and 13 healthcare professionals. Interviews were recorded on digital audio and analysed using an inductive thematic analysis within a realist framework.

Result: Informal caregivers discussed five themes of support needs relating to being overburdened by administrative demand, in contact with healthcare providers, providing caregiving at home, especially during the terminal phase, and having or lacking social support. Healthcare professionals discussed three themes of support needs of informal caregivers which related to the general institutional support for informal caregivers, their own work as caring for informal caregivers, and the challenges in healthcare for families with ALS they encountered.

Significance of results: Informal caregiving for people with ALS can be demanding. This study provides evidence for improvements in supporting informal caregivers. It shows administrative needs of informal caregivers, stresses their needs regarding advance care planning early in the context of ALS, and underlines the importance of social support, be it in peer-groups or community care.

Keywords: informal caregiving, supportive needs, amyotrophic lateral sclerosis, palliative care, qualitative study

Introduction

Amyotrophic lateral sclerosis (ALS) is a terminal, neurodegenerative disease leading to progressive immobilization and paralysis. Due to its fatality, people with ALS (pwALS) need intensive and complex care. This essential care is most often provided by family and friends, the *informal caregivers of pwALS*.

Due to the high burden of care for informal caregivers (de Wit et al., 2018) supportive needs, e.g., related to healthcare service use (Brown, Lattimer, & Tudball, 2005), have been studied in the context of ALS (Poppe et al., 2020; Oh & Kim, 2017). Informal caregivers and patients form a distinctive caregiving dyad, and personal characteristics and relationship dynamics play a role in how needs form out (Oh & Schepp, 2013). This is the first study of this kind in Switzerland. As cultural context is important in end-of-life care (Gysels et al., 2012), highlighting needs within the Swiss context is important. This context includes political and legal factors influencing caregiving and the availability of financial and personal support, and specialist palliative care as well as funding of auxiliary aids after transition in retirement age (Rickli, 2016).

To the existing corpus of knowledge available on informal caregivers' needs, this paper adds new information using interview data. The paper asks what supportive needs informal caregivers of pwALS have from the perspective of informal caregivers and healthcare professionals supporting families with ALS. We understand needs as capacities of informal caregivers to benefit from support (Asadi-Lari, Packham, & Gray, 2003).

Methods

Sample

The sample consisted of 9 current caregivers, 14 bereaved informal caregivers, and 13 healthcare professionals. The healthcare professionals included five physicians (palliative or hospice care, neurology, and general practice), three physio- or respiratory therapists (ALS clinic and private practice), and five specialised nurses (neuromuscular centres or private practice) providing care to individuals with ALS and their families. Informal caregivers were mostly family caregivers, with an age range from 28 to 74 years. From the family caregivers, most (96 %) were spouses or daughters of individuals diagnosed with ALS. Of the 23, nine informal caregivers were currently providing care to a pwALS. Informal caregivers were recruited purposefully through ALS clinics in the German-speaking part of Switzerland, through patient initiatives (Verein ALS Schweiz), peer contacts of former caregivers, and snowball sampling.

Informal caregivers contacted the interviewers via phone, mail, or email after having heard of the study through healthcare providers and handouts in peer-groups. With informal caregivers, interviews took place mainly in person in the participants' homes to accommodate time and caregiving constraints. Healthcare professionals were mostly contacted in advance and interviewed via telephone. Interviewees were comprehensively informed about the study aims during informed consent procedure. In two interviews with current caregivers, pwALS were present and participated in the interviews. None of the interviewees dropped out of the study or refused to participate.

Interviews were conducted and audiotaped after all participants had provided written informed consent. Interviews were between 25 – 130 minutes in length and interviews with healthcare professionals were typically shorter than interviews with caregivers. Each participant was interviewed only one time during the time of 2018-2020. Interviews were transcribed verbatim in Standard German using MAXQDA (Verbi Software, 2020). All quotations included in this article were translated to English from Standard German.

The interviewers were a female physician (MD) working in general practice and a male clinical psychologist (MSc.) working in the academic setting. Both had prior practice in conducting qualitative research interviews or analysis and were repeatedly trained in qualitative data collection. The first four interviews served as a pilot for the semi-structured interview guide and the interviewers' interview skills, which were supervised by a senior researcher. For informal caregivers, the interview guide comprised of questions regarding practical supportive needs during caregiving, difficult situations, advance care planning and end-of-life, as well as the time after bereavement (for more information see interview guides for all groups in the Appendix). For healthcare professionals, the interview guide centred on support for caregivers. The interviewers did not have any prior relationship to the research participants, nor did the participants have specific knowledge about the interviewers apart from general information about the project.

Analysis

We conducted an inductive thematic analysis within a realist framework (Braun & Clarke, 2006). Correspondingly, we approached this by inductively coding segment by segment of the interview data. Segments consisted of units that relayed a meaning relevant for the context (usually one to three sentences).

Data analysis was performed in parallel to data collection. Data saturation was monitored while inductively coding and achieved when recurrent coding occurred. Data saturation was therefore understood as post-hoc information redundancy achieved during data analysis (Braun & Clarke, 2019). Overall, 20 interview transcripts were coded by hand by teams of two to three coders (six coders overall) to satisfy inter-coder agreement of approach to the data. The remaining interviews were coded inductively using MaxQDA solely by the first author. Inductive codes were grouped by themes and subthemes. In the results section, we present the results from the analysis of interview of informal caregivers and healthcare professionals in two parts to provide a comparison of themes.

Ethics approval

The project was submitted for review by the cantonal research ethics committee (Ethikkommission Nordwest- und Zentralschweiz, EKNZ) through swissethics. As the project does not qualify as research with human subjects under Swiss laws, a certificate of non-objection was issued by the EKNZ. Despite not being subject to human research laws, we felt that it was necessary to uphold the standards required by the same laws of Switzerland, that is, having written informed consent of every participant, as well as storing data separately in a de-identified manner. Furthermore, no identifying information is presented in the paper that could jeopardize confidentiality.

Results

Informal caregivers

Informal caregivers discussed five support needs in the following tasks, situations, or places, (1) *administrative demand; (2) healthcare providers,* (3) *at home*, (4) *during the terminal phase,* and *(5) social support*.

Theme	**Quote**
Administrative demand	1. “We needed an alarm system for my mother / this was actually rejected twice although it would have been clear that she needed something like that. Then we actually wanted to write them again and again but then it has actually // so the disease was so advanced that we had to look after her for 24 hours anyway.” Participant 6217 2. “Not all but so yes, it annoys me that the insurance is so stupid because of a bed or that the health insurances do not even want to pay for any medication or for speech therapy, although they know that speech therapy is central. .... Despite the third request for reconsideration, they say no, ... And the whole paperwork is annoying.” Participant 9059“ 3. And even to know that there is no other way. I then took advice from a lawyer / um there is no way that one can somehow get off work to care for relatives or get a vacation for some time or so. I found that stressful.” Participant 9352
Healthcare providers	4. “And then this family doctor, I could really call him whenever I wanted. So I was scared around Christmas when everyone was on vacation and he would have pronounced dyspnea, I got morphine from him with the instructions, but I said, I'm scared, I'm scared of this morphine. He said in any case I am from Christmas to New Year we won't go away. And said you can call me at any time....” Participant 6697 5. „I was there. Yes, exactly. We were pretty offended, so she was just told, ‘You have a fatal diagnosis. You have three to five years to live. Do whatever you enjoy now, now. If you have any questions, you can call me again. Goodbye’” Participant 5450. 6. „Shortly after the diagnosis and there we really had, I would say, a three-month paralysis in shock that was very, very difficult to break through, you just want to protect yourself.” Participant 8807. 7. „I’ll give you an example now: I had to take my husband to the rehabilitation centre on Monday. From (hospital) to (rehabilitation centre), I brought him there. On Wednesday, when I came, I said to him: ‘You, has / has someone already shaved you?’. He couldn’t do it himself anymore.” Participant 0735

	8. „I could / I didn’t really want to, uh, go to the meetings (peer meetings) at the beginning because I was afraid that, I, the other, would meet patients and I was scared to be honest (to face the diagnosis). But at some point, I did / it got better over time and then I signed up for that afternoon and it was really good.” Participant 4109
At home	9. “But now the move, you will be torn out of your familiar social environment, of course you will lose one or the other relationship, that could have offered you the support.” Participant 4673 10. “I think that was also very important for my mom. She was then able (due to continuity of care) to build trust in this person and that was like not every time someone else to whom had you to tell everything again.” Participant 9352
Support during the terminal phase	11. “We had already programmed the one with the computer people, we already had put together the most important like ‘The arm hurts me’, ‘The back hurts me.’ ‘Can I have a drink?’. Simple so the most important ones that he uses to create a program so that we know, ‘1’ means this and that. That wasn’t done yet when he died. But it was prepared because we wanted to have if it no longer works.” Participant 6697 12. “I then also printed out information about options and gave my mother that she also knew that what it was and that if she wanted to put an earlier end to her life, that she could do it as long she could still sign the document. It is difficult because you have to give yourself a pill. Well that wouldn’t have worked anyway. She wasn’t very decisive about such things.” Participant 2504 13. “Then I registered my wife (at the assisted dying organization) on the computer which went without any problems, but later that has to be made by the patient himself and not a relative. But then it was all for nothing when the diagnosis of dementia came and then assisted dying is impossible. I knew that. Then I also knew that at some point she would no longer be able to eat independently and the we thought about another option to assisted dying, no more nutrition. And then I wanted to know about this from the family doctor what was going on. If you take this option – I actually didn’t really hear what I wanted to hear – so I paused a bit. I informed myself about other options, and well, starvation is a relatively hard way to die. It is not painless. The

	consequence was that in consultation with the hospital, we had the PEG and the catheter done at the same time." Participant 4673 14. "With the shortness of breath, this woman (name palliative care team) already had everything at the beginning, gave morphine droplets and instructed us what to do then. And so, um, so um like lollipops that you put in your mouth on the mucous membrane. And when it got worse, she also had the syringes, she had everything she then prescribed how I was allowed to inject it and I always wrote everything down exactly. And she really came by every day for about three weeks, every day. And of course that is great, then you could observe the situation together." Participant 3719
Social and other support	15. "Yes, I think we have a very good environment, that is certainly very important, or neighbors, family. That you are simply well embedded, right? I think that brings a lot." Participant 6598 16. "Because we need to hear "You, we have to go through it now, somehow we have to go through this now and we can do it", but I think you could do a lot more. I don't know I always felt very comfortable after we stay with (name of doctor) because she said "You did well and just keep doing it this way". Participant 6729

Table 1: Themes of supportive needs of informal caregivers

Administrative demand

Informal caregivers highlighted the support needs with administrative demands. Vital was the importance of information about administrative tasks at hand, about available support options, being referred to the responsible institutions, and to have someone else perform administrative tasks.

A source of need were problematic interactions with insurance providers. During working age, informal caregivers and patients in Switzerland have in principle access to invalidity insurance which allows them to access different lines of support. Caregivers noted regional differences and a need for openness and the willingness of services providers to support them. PwALS could get support through disability reimbursement which in turn provided the informal caregivers with additional respite help and financial aid. However, disability was assessed momentarily and only reassessed once a year. This means that help came sometimes too late for patients and informal caregivers (Table 1, quote 1).

For insurance, especially the transition from working to retirement age came as a predicament for caregivers. Many privileges associated with invalidity insurance were lost with old age insurance. Generally, informal caregivers viewed administrative work metaphorically as a fight, where they needed support (Table 1, quote 2).

Another source of need was work obligations; flexible working hours, an understanding employer, and supportive colleagues are needed. Self-employed informal caregivers had trouble finding people able to replace their positions. The lack of legal possibility to reduce working hours was mentioned by some informal caregivers, while others resolved this by taking sick leave (Table 1, quote 3).

Healthcare providers

Informal caregivers mentioned several needs in relation to physicians, e.g., a need for a caring physician, especially during the diagnosis communication. Moreover, they expected engagement in the treatment and acting as a patient advocate. Especially family physicians were expected to perform this role, working as a contact point in the healthcare system (Table 1, quote 4).

However, some informal caregivers complained about the lack of information and empathic support following a diagnosis of ALS from physicians (Table 1, quote 5). Active engagement of physicians to coordinate care, ensure transfer to specialist centres, and clear communications with informal caregivers regarding time needed to deal with the diagnosis and coping in general were mentioned as important to have (Table 1, quote 6). Furthermore, informal caregivers mentioned that they needed support when pwALS avoided facing the diagnosis and its fatal implications.

When pwALS were hospitalised, some informal caregiver experienced patient-related needs which were not completely met (Table, 1, quote 7). Here, informal caregivers filled the essential gaps. Generally, healthcare professionals were needed to coordinate care and provide case management. Specialised support came through specialist nurses providing information, counselling, coordination, instruction, and key contact.

Instructive modular care trainings were provided by them to informal caregivers every three months dealing with a variety of skills. Due to the modularity, informal caregivers sometimes missed certain training. For example, the progression of ALS was quicker than the availability of care trainings. Another problem was the emotional toll of these trainings (Table 1, quote 8).

For informal caregivers, proximity to ALS centre was of advantage. As pwALS becomes more immobile, the harder travel to ALS centres becomes. Hence, telephone support was highly valued.

At home

Most of the care for pwALS took place at home and it must be adapted for that purpose. Informal caregivers discussed needs to make the home fit to provide care. There, support from architects was needed who knew about ALS and the progredient disability. A lift and wheelchair-ready cars were need for mobility. If they are unable to do these changes in their homes, they had to move. Moves are difficult because social support is then lacking (Table 1, quote 9).

At home, informal caregivers reported having and needing care assistants to provide them respite and difficulties finding experienced assistants. They communicated a lack of respite structures at home or outside. One of the most vital needs at home were support with specialized care aids to keep up with the deteriorating condition of pwALS. Often informal caregivers organized care aids too late, i.e., by the time equipment was at home, the health of pwALS had changed so that the equipment is no longer needed or appropriate (e.g., wheelchair to electric wheelchair). Hence, informal caregivers expressed a need for a central depot of care aids that would address the changing needs of pwALS, as well as being able to get unbureaucratic financial support even if the care aids would only been used in the future. However, these kinds of depots were already used by other informal caregivers.

Competent homecare services were valued support but were often limited to a short period. For instance, homecare nursing support only came in the mornings and afternoons, while informal caregivers would require readily available round-the-clock care. Continuity and flexibility of care were both much desired, but often unmet (Table

1, quote 10). Due to immobility, therapies accessed by pwALS were increasingly delivered at home. Informal caregivers needed therapists and physicians to come to their home but only a part of physicians and therapists provided such services.

Support during the terminal phase

For support in the terminal phase, informal caregivers highlighted the need to plan care in advance. This encompasses early organization and information as well as the opportunity to do so in the institutional context.

Informal caregivers needed help with communication in the later stages. While in their private life communication could often be established by family caregivers and pwALS, in the healthcare systems problems were encountered when healthcare professionals equated communication incapacity with stupidity or dementia. Here, information and training for involved personnel was considered as a need as well as early integration and calibration of electronic communication devices, including integration with ventilation devices (Table 1, quote 11). Generally, ventilation was a situation of need for caregivers with patients being afraid of dying of dyspnoea pointing to a gap of information about palliative care. There were problems with non-invasive ventilation, especially when pwALS could not remove the mask.

A key mistake that bereaved caregivers spoke about is planning too late. Informal caregivers needed earlier and clear communication of possible medical interventions and end-of-life options as well as of support with prognostic uncertainty. Problems were the decision-making capacity as well as when pwALS left it to the family caregivers to decide or were not decisive (Table 1, quote 12).

The end-of-life was characterized by increased need for support. Informal caregivers were significantly involved in decision-making surrounding decisions that pwALS had protracted during their illness like insertion of a feeding tube, and invasive ventilation, as well as in end-of-life decisions (e.g. deep sedation, and termination of ventilation). As many of the decisions rendered by pwALS hinged on the provision of care at home, informal caregivers were key and often decided together with or in place of pwALS (Table 1, quote 13). Here, they needed time to weigh all the options and get support through specialist palliative care teams (Table 1, quote 14). During these decisions, feeling supported and not alone was vital. If the place of death was not the home, some informal caregivers reported having a hard time finding a suitable institution to support pwALS at the end-of-life.

During the end-of-life stage, informal caregivers report emotional needs. Thus, early access to psychological and spiritual care were necessary support needs, as well as the activation of resources needed to provide care.

Social and other support

Social support from other family members and friends was very important for informal caregivers. Their social network needed to have knowledge of ALS' intricacies, and being able to accept ALS. Often, informal caregivers had problems if their friends feared ALS and withdrew from contact. In their communities, informal caregivers valued if neighbours helped them out (Table 1, quote 15).

Societal factors played a big role; while informal caregivers noticed that knowledge about ALS has improved in the last 15 years, there was not extensive knowledge even among healthcare professionals. Informal caregivers valued if healthcare professionals recognized the work of caregivers (Table 1, quote 16).

When asked directly, informal caregivers remarked that they in general need more holistic support, being met where they stand regarding ALS, someone to talk to, specific situations of care (nightly toilet walks), friends for moral support and someone to take the initiative to care for them.

Healthcare professionals

There are three themes delineating support needs of informal caregivers from the analysis of the interviews conducted with healthcare professionals: *Institutional support for informal caregivers, caring for informal caregivers*, and *challenges in healthcare for families with ALS*.

Theme	**Quote**
Institutional support for	1. „Yes, it's a very intensive consultation, activity and ahm accompaniment, often also very emotional for everyone involved because it is a life-limiting and sometimes very rapid disease that

informal caregivers	more or less cuts into all areas of life, but it is a job that just because of the way it is, is existential. Hand in hand there is an appreciation, so most of the patients and relatives are very cooperative and grateful for the advice and support services and the network that we offer." Participant 231 2. "It depends on early palliative care. Actually, the ALS is an almost paradigmatic disease for palliative medicine. Now, of course, one could argue that the patient needs a palliative team from the initial diagnosis. I would have argued this way earlier in my professional life. But I actually find the system as it is now very useful. Namely, that a large part of palliative is general palliative care provided by the professional caregivers, who are neurologists for example, and that we do not have to see everyone. We cannot see all, for resource reasons. But that we then come into play when it, it is difficult and complex. So, I find, this more modern model that is needs-adapted makes much more sense than what I preached myself earlier." Participant 810 3. "We wanted to do this low-threshold, help for self-help for relatives, meetings, courses or just coffee meetings, that is something that we wanted to do but we simply do not have the resources to rent a large table in a café. Where relatives just empty their heads and talk about their garden or whatever. In the peer group. These offers are completely missing." Participant 167 4. "It is very difficult to find places for ventilated patients here, including quadriplegics, very difficult. A quadriplegic might go home again and be cared for by his family, for example in (institution), there are ventilation places in (town) but these are really few and with the ALS patients, the disease simply progresses." Participant 874
Caring for informal caregivers	5. "So I think it is typical that the family, the relatives, suffer more than the ALS patients themselves. The ALS patients are actually very comfortable in therapy, how should I put it, compliant, who work very well with, are very well motivated, or are. And, and the relatives, the carers, they see the situation a little differently, right? They see better where it's going." Participant 816

	6. “Then I go to them at home, then that is half an hour longer and that’s half an hour longer unpaid and that’s on me. And not every day because I really need time and then there are statements like ‘I don’t want to live anymore’ when people notice that there is no relief.” Participant 475 7. “Yes, let's put it this way, at the beginning a lot was learning by doing, these were exotic patients, as I said, we did not do them justice at all, the psychosocial care was not an issue at all - as with many other patients - and over time I simply gained experience through these many years, I've read a lot, so I did it myself, I go to congresses, sometimes where that's the topic, so that's how I continue my education, I think that's important, ah, yes. But I don't have a structured training specifically for ALS and the advance care planning was also developed by palliative medicine specialists over the years, so we are the ones who have established that, that is learning by doing in our field.” Participant 167 8. “Well. If the situation of talk about dying arises, then we have the opportunity to talk about it yes. And you hear, well, I am not from Switzerland, my problem is that here basically hardly anyone uses the word “dying”, yes that is the bad, bad word one rather talks about falling asleep. And if the relative doesn’t want to listen and then falling asleep means falling asleep but not death, yes? So really you don’t have to say it angrily, yes, but you have to say it that everyone understands and can also hear it. Even if they may even say they don’t want to hear.” Participant 319
Challenges in healthcare for families with ALS	9. “That already went bad, high symptom load then we also created a network with physician (name) and he first went to her, and then he was in a network which relieved the family. It was already very late in the disease, and he then came here with his wife.” Participant 167 10. “The biggest challenge I think is frontotemporal dementia which people develop but they remain capable of judgement. Effectively getting problems in behavior, where it gets very difficult for the relatives. Where it is sometimes more stressful than anything else, where you can’t progress in counselling.” Participant 344

	11. “I actually once experienced with a patient who was flown in as an emergency, intubated from another hospital, which just had no clue about ALS, ehm and came intubated was then also in intensive care, … and then tracheostomized. He was still young enough, eh, we could still teach him to handle everything himself with his tracheostomy, with his ventilation, with the suction device, but we had the specific problem that the wife didn't want to know anything about it. Because she was still so overloaded and burdened with the whole situation that / That we weren't allowed to involve her, yes, simply to spare her.“ 319 12. „Interviewer: Then maybe finally: What kind of support needs do you yourself have in your work with ALS patients and their relatives? Participant: supervision. Yes.“ 115

Table 2: Themes of support of healthcare professionals

Institutional support for informal caregivers

Healthcare professionals discussed the importance of support offered to informal caregivers, by themselves and other professionals. Depending on their professional role, they described their professional responsibilities. Specialized nurses reported a variety of general task and being involved from diagnosis. They also reported visiting families at home, planning and coordinating care at home and providing case management. Healthcare professionals provided care within a network (Table 2, quote 1).

A valuable source of support for informal caregivers are assistants for daily help. Healthcare professionals highlighted the need for specialized medical care; this encompassed early integration of palliative care, adapted to the needs (Table 2, quote 2).

Generally, healthcare professionals reported how support of informal caregivers depended on the demand for and acceptance of support. Professional caregivers noted how support for financial and administrative needs was often lacking, especially regarding social insurances.

Support offers identified by healthcare professionals ranged from organizing care aids and appliances, instructing, and supporting involved care teams, and organizing respite services. They point to the need for more volunteers to provide respite care, peer groups and self-help groups in the context of ALS (Table 2, quote 3).

The end-of-life was marked as a crucial phase of support. Healthcare professionals mentioned different lines of support for informal caregivers and pwALS (mobile palliative care, hospice care, assisted dying), but also lack of institutions for long-term care, especially with invasive ventilation (Table 2, quote 4).

Caring for informal caregivers

Professional caregivers from multiple disciplines stated that support for informal caregivers started when first meeting the family, often after diagnosis. Healthcare professionals highlighted the burden informal caregivers had to deal with as they saw the disease progress (Table 2, quote 5).

Support through the disease course, advice and counselling were identified as key task where professional caregivers met needs of informal caregivers. Furthermore, professional caregivers revealed their role as advocates of patients and families, as well as providing recognition of the work done by informal caregivers. Being trusted by informal caregivers was central in caring for them.

Healthcare professionals highlighted the fatality of ALS and the need for a professional role understanding, encompassing both compassion and professional distance. They reported doing unpaid work because they needed time for extensive counselling (Table 2, quote 6).

Most professional caregivers reported having learned to support patients and families with ALS by "learning by doing". While some had extensive training before coming to support families, they also noted how the novelty of support for ALS required constant training, education, and information (Table 2, quote 7). In turn, professional caregivers informed, trained, and instructed informal caregivers and thereby meeting their needs.

The care the professionals provided which addressed the supportive needs of informal caregivers was orientated towards the resources of informal caregivers and was planned out proactively in advance. Especially decision-making and planning in the later stages was a key activity where professional caregivers supported caregivers. Other supportive activities included planning and coordinating medical care, setting up advance directives, preparing the caregivers and pwALS for impeding death by talking frankly with them (Table 2, quote 8). Professionals regarded the possibility to visit families at home to observe the situation as important, as well as the advantage of continuously providing care for pwALS.

Challenges in supporting families with ALS

Healthcare professionals identified a range of challenges when supporting family caregivers and pwALS. In itself, working with the difficult diagnosis of ALS was perceived as burdening as well as a learning experience from different ways of coping with a fatal illness. Most often a first challenge was when the contact with the families came too late, and there was severe disability and an inability to plan ahead (Table 2, quote 9).

Neurocognitive and behavioural impairments of pwALS were noted by healthcare professionals as particularly troubling for informal caregivers (Table 2, quote 10). Healthcare professionals remarked how neurocognitive impairment might influence the caregiving relationship.

The end-of-life gave rise to challenges and moral dilemmas. Dying and planning in advance for death were viewed as different depending on the cultural background. Ventilation, both non-invasive and invasive, was a key issue (Table 2, quote 11). Professionals noted problems with mask ventilation in ALS and most notably, problematized invasive ventilation by tracheostomy in the restricted healthcare context of Switzerland. Care at home until death was viewed as costly and burdening, where round-the-clock care was needed and had to be organized, as well as the implications of high symptom load for all involved were discussed. Professionals viewed communication at the end-of-life as a key challenge within palliative care. In contrast to palliative care in oncological patients, palliative medicine in the context of ALS was viewed as more complex and symptomatic treatment as difficult to control at times. Wishes to die and wishes to hasten death portrayed situations where professionals reported having difficulty.

Generally, challenges reported by healthcare professionals were the voluntariness of caregiving, as well as the gendered nature of caregiving in ALS with women providing the majority of care, and the problems arising from societal challenges such a lack of financial resources in the healthcare sector. Additionally, financially burdened families were a challenge for professionals. That is, they underlined that fact that for families, financial support was missing to pay for care aids and appliances as well as respite workers. From the perspective of institutions, resources for home care professionals, home visits, and night support were lacking. For their own work, professionals problematised interdisciplinary cooperation and the lack of supervision (Table 2, quote 12).

Discussion

Both interviews with informal and professional caregivers stressed the need for early care and advance care planning, depending on the needs and resources of the families. Both groups also converge on the idea of the importance of social support, be it in peer-groups or community care. The value of support through peers is a common finding, e.g., in a recent study by de Wit and colleagues (2019).

From both caregivers and healthcare professionals, administrative demand is seen as a need where caregivers would appreciate support, especially when such support could result in avoiding time-delayed resources, financial and otherwise, offered by the welfare state. De Wit et al. (2019) write for the Dutch context that this work "is generally time-consuming and bureaucratic, whereas the need is often urgent" (p. 195) with which our data concur.

The home as a central place of care brings with it specific supportive needs. The decision to care at home until death of pwALS is highly depended on factors such as knowledge of and skills in early advance care planning including emergency plans tailored to the disease and individualized goals of care, the availability of respite care and palliative care support when needed, as well as informal caregivers being able to take the time to provide such care. These results map well onto the existing qualitative data (O'Brien et al., 2011; Ushikubo & Suzuki, 2015; Nakai, Narita, & Tomimoto, 2017). Professional caregivers offer home visits, but also point to the difficulty to finance such visits and to finance complex palliative care at home. Similar to the study by Galvin and colleagues (2018), external support and services and patient-related behaviour played a role in both informal caregiver and healthcare professionals report. Psychological support was not mentioned as a need by informal caregivers in our study. Given the absence of evidence for psychosocial interventions (de Wit et al., 2020) and the lack of time caregivers are faced with, this provides evidence that psychological support might be only needed in specific circumstances, e.g. complicated grief in bereaved caregivers (Aoun et al., 2020).

Our results have implications for provision of care and support for informal caregivers in Switzerland. One is that the role of family physician is crucial. Alvarado and Liebig (2015) show that general physicians lack opportunities to acquire the needed skills for palliative care. For ALS, these skills need to include advance care planning and serious conversations around severe illness and death. However, as general physicians might only see few ALS patients, they may act more as an intermediary and contact person and bring in specialist palliative care.

Future research on palliative care in Switzerland is needed (Reeves, Schweighoffer, & Liebig, 2021). Financing and educational opportunities for palliative care are highly varied in the Swiss cantons. The Swiss national palliative care strategy 2010—2015 has improved the situation of caregivers as shown by the availability of mobile palliative care services supporting some informal caregivers in this study. Low-threshold psychosocial interventions for informal caregivers demand more research. While informal caregivers valued ALS-specific peer group meetings, infrequency of these meetings gives rise to a need for continuously available support. Volunteers trained in palliative care (Claxton-Oldfield, 2015), ideally with experience in ALS, might be an example of low-threshold support that could be organised regionally. Additionally, the availability of professional respite support seems crucial to enable caregiving.

Healthcare professionals highlighted different challenges for informal caregivers, notably neurocognitive impairments, and implications upon the caregiving dyad. This issue was not brought up by informal caregivers. It is possible that these are implicit factors in relationship which are easier viewed from the outside, or that the informal caregivers involved did not have or want to voice these issues. It might also be that healthcare professionals have learned to recognize signs of neurocognitive impairment, while family caregivers normalize impairments. This demands more research as there is evidence that behavioural impairments particularly relate to the subjective burden of caregiving (Merrilees et al., 2010).

Finally, this study adds to the literature on needs of informal caregivers of pwALS and the need to tailor interventions to each unique family system. Some informal caregivers brought up privacy concerns for external help, while others would have been most thankful for it. Hence, supporting informal caregivers for pwALS needs to be highly adapted to their needs.

Limitations

This qualitative study is necessarily limited, as our data are not generalizable. Caregiving is a context-dependent activity (e.g., cultural background, welfare-state support), and these results should be viewed within the Swiss context. Additionally, the participants were sampled by purposive sampling, and this necessarily selects for informal caregivers who felt able to give an interview (e.g., where not overburdened or amidst severe grief). Furthermore, while our interview questions were open and varied, social desirability might have played a role in the interviews (e.g., refraining from talking about socially undesirable feelings towards pwALS).

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Chapter 5.

“Walking a tightrope”: A qualitative study on informal caregiving in palliative care for amyotrophic lateral sclerosis

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"Walking a tight rope": A qualitative study on informal caregiving in palliative care for amyotrophic lateral sclerosis

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Abstract

Informal caregivers, mainly family members and friends, provide supportive and palliative care throughout for people with ALS (pwALS) during their terminal disease course. In this study, we provide a theoretical understanding of informal caregiving in ALS using a grounded theory approach. The data stem from 23 semi-structured interviews conducted with informal caregivers of people with ALS in Switzerland. Due to the expected death of the care-recipient, our grounded theory approach outlines informal caregivers' caregiving work as an effort to secure a balance among different caregiving activities, which feed into the final stage of providing palliative care. Overall, our theoretical understanding of ALS informal caregiving work encompasses the core category, holding the balance and four secondary categories: organising support, being present, managing everyday life, and keeping up with the illness. The core category, holding the balance aims at ensuring care and normalcy as disease progresses and until the end-of-life. On this understanding, those caregivers that succeed in holding the balance can provide care at home until death. Holding the balance is heavily influenced by contextual factors of caregiving, or activities of caregiving where the goal is to ensure the quality of life of the pwALS. As there is a heterogeneity of speed and subtype of progression of ALS, our work accounts for multiple caregiving trajectories.

What is known about this topic and what this paper adds?

- Informal caregiving for people with ALS is a demanding and variable process
- Using a grounded theory approach, informal caregiving in ALS is seen as a constant endeavour to hold the balance between different sub-activities (organising support, being present, managing everyday life, and keeping up with the illness)

- This theoretical understanding provides a flexible approach to supporting caregivers during caregiving for people with ALS

Keywords: informal caregiving, amyotrophic lateral sclerosis, grounded theory, family, palliative care

Introduction

Amyotrophic lateral sclerosis (ALS) is a terminal neurodegenerative disease leading to progressive muscle weakness caused by genetic and environmental factors (Al-Chalabi et al., 2014). Given the incurable nature of ALS, any care provided to people with ALS (pwALS), whether by formal or informal caregivers, can from diagnosis be understood as palliative according to the definition by the World Health Organization (Sepúlveda et al., 2002). ALS is often described as an illness affecting the whole family, as it takes its toll not only on pwALS, but also on their immediate social environment (Cipolletta & Amicucci, 2015; de Wit et al., 2019). Informal caregivers, mainly family members, provide supportive and palliative care throughout the disease course (Aoun et al., 2012). During the caregiving period, informal caregivers experience different needs such as external support and assistance, psychological-emotional support, and patients' acceptance of available services (Galvin et al., 2018) as well as emotional needs concerning fears for their own future and the patient's (Galvin et al., 2016). Informal caregivers fear that each crisis could mean the end and worry about the future. Hence, Aoun and colleagues (2012) concluded that the work of informal carers is demanding.

Lerum and colleagues (2016) identified five lines of work of informal caregivers: *immediate care work, seeking information and clarity, managing competing obligations, maintaining normality*, and *managing external resources and assistance*. Their study follows a sociology of work of informal caregiving for and coping with chronic illness, which comprises three key works according to Corbin and Strauss (1985): *illness work*, *everyday life work*, and *biographical work*. Instead of focusing on illness experience, the authors used the concept of *illness trajectory* which "refers not merely to (1) the course of an illness, but also (2) to all the related work, as well as (3) the impact on both the workers and their relationships that (4) then further affect the management of that course of illness and the fate of the person who has it" (Corbin & Strauss, 1985, p. 225).

However, this trajectory model does not fully explore the work of informal caregiving, especially in an expected death (Penrod et al., 2011) model, such as in ALS.

In this paper, we seek to depict informal caregiving for pwALS as their disease progression continues towards palliative care. Our analysis is grounded in 23 semi-structured interviews conducted with informal caregivers of pwALS. More specifically, our research also addresses what lines of work informal caregivers perform to enable caregiving at home – and how the caregiving trajectories differ according to place and time of death.

Methods

We used a qualitative approach based on the constructivist grounded theory method (Charmaz, 2006) which focuses on reported processes and actions of informal caregivers to build a theoretical understanding of informal caregiving in the context of palliative care for pwALS. Key to this approach were intensive line by line initial coding, memo writing, focused coding and the definition of categories.

Sample

Our sample comprised of 23 informal caregivers, mostly family caregivers, with an age range from 28 to 74 years. From the family caregivers, most (96%) were spouses or daughters of pwALS. Informal caregivers were recruited purposefully through ALS centres in German speaking part of Switzerland, through patient initiatives (ALS Verein Schweiz), peer contacts of former caregivers and via snowball sampling. Theoretical sampling, while an important pillar of a grounded theory method (Charmaz, 2006), was not possible as the participants for the phenomenon under study were not widely available. Hence, we could not choose participants along theoretical lines. However, as the interviews resulted in rich data and densely coded text, and focused coding resulted in a dense description of the material.

Participants contacted the interviewers via phone, mail or email after having heard of the study. The interviews took place between 2018 and 2020 mainly in person in the participants' home to accommodate time and caregiving constraints. Interviews were conducted and audiotaped after all participants had provided written informed consent.

Interviews were between 30 – 120 minutes in length. Each participant was interviewed only one time. Interviews were transcribed verbatim in Standard High German using MAXQDA (VERBI, 2020).

The interviewers were a female physician (MD) working in general practice and a male clinical psychologist (MSc.) working in the academic setting. Both were native Swiss German and German speakers, had prior practice in conducting qualitative research interviews or analysis, and were repeatedly trained in qualitative data collection. The first four interviews served as a pilot for the semi-structured interview guide and the interviewers' interview skills, which were supervised by a senior researcher. The interview guide comprised of open-ended questions regarding practical supportive needs during the caregiving trajectory, difficult situations caregivers had experienced, advance care planning and end-of-life experiences, as well as the time after bereavement. The interviewers did not have any prior relationship to the research participants, nor did the participants have specific knowledge about the interviewers apart from general information about the project.

Data analysis

Coding was performed in parallel to data collection. As the goal was to gain a theoretical understanding of the process of caregiving, codes were written in the gerund form. Coding was an iterative and interactive process. Initially, five interview transcripts were coded by hand in teams of two to three coders (six coders overall) to satisfy rigor and to get a common understanding of the initial coding process. While initial and focused coding, memos were written for the category building part of theory building. After initial coding, initial codes were coded in a focused manner. Theoretical saturation was monitored during the initial and focused coding phase, and assumed when no new focused codes could be constructed. Together with the focused codes, memos were used to construct five categories of the informal caregiving process in ALS disease course, especially with regard to caregiving in palliative and end-of-life care. Finally, categories were translated from German to English.

Ethics approval

The project was submitted for review by the cantonal research ethics committee (Ethikkommission Nordwest- und Zentralschweiz, EKNZ) through swissethics. As the project does not qualify as research with human subjects under Swiss laws, a certificate of non-objection was issued by the EKNZ. Despite not being subject to human research laws, we felt that it was necessary to uphold the standards required by the same laws of Switzerland, that is, having written informed consent of every participant, as well as storing data separately in a de-identified manner. Furthermore, no identifying information is presented in the paper that could jeopardize confidentiality.

Results

Due to the expected death of the care-recipient, our grounded theory approach outlines informal ALS caregivers' work as an effort to secure a balance among different caregiving activities, which all feed into the final stage of providing palliative care. This balance is characterized by the core category *holding the balance,* an organising element to the other categories. This means that the other categories of caregiving are all directed towards this core category. Overall, our theoretical understanding of ALS informal caregiving work encompasses the core category, *holding the balance,* and four secondary categories: *Organizing support, being present, managing everyday life,* and *keeping up with the illness.*

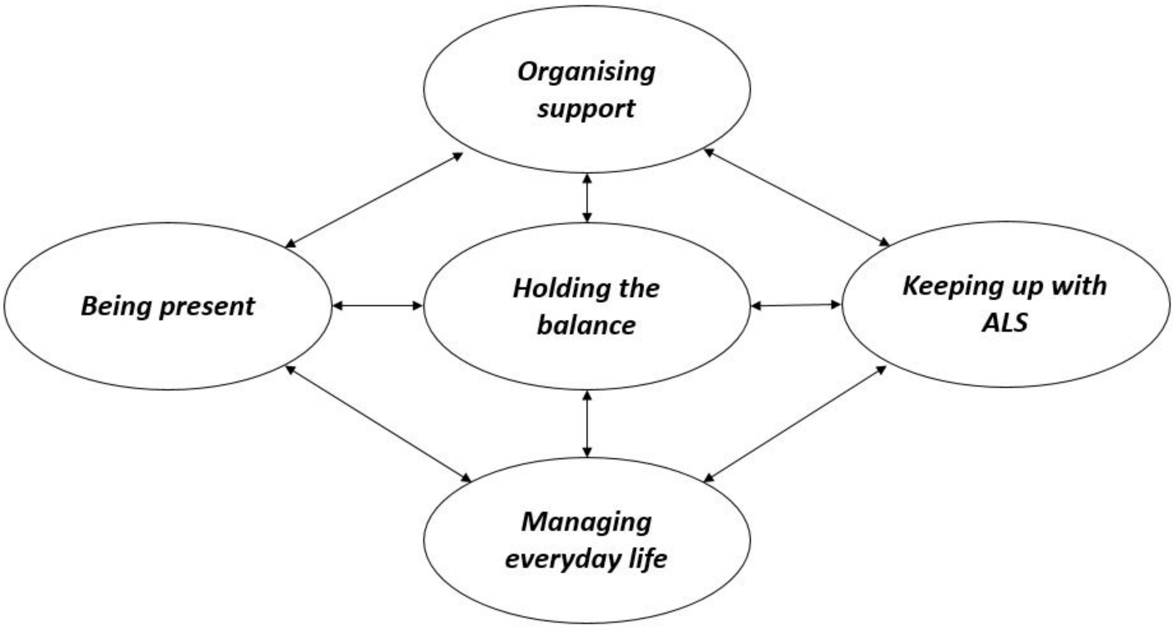

Figure 1: Five categories of informal caregiving in ALS

Holding the balance

Generally, from the diagnosis of ALS informal caregivers worry about the balance in their lives to care for pwALS. Holding the balance is the core category encompassing all the other categories of work that informal caregivers perform such as balancing family life and caregiving.

> Well, I have this always in sight and well it is difficult: to find the balance between my son and my husband, the rest of the family and also myself. (current female spousal caregiver, 6009)

Another participant noted the need of not only balancing family obligations but also keeping up one's own strength in caregiving as an effort to maintain the balance.

> We are well prepared. My family physician is the same as my wife's, but she / she supports the whole family. And she says that I need to take care of myself, that I keep my strength and notice / like in sports when you need extra support. It is a balancing act, well, that's what one notices. (current male spousal caregiver, 7777)

Crucial for the task of holding balance is the relationship with pwALS, as well as distinct personalities of both the carer and the cared-for. Using the metaphor of a power bank, one caregiver remarked how she was able to go through the caregiving process despite it being tough:

> What actually helps me is the positive nature of the sick person. That's my power bank (laughs), that's the way it is. (current female spousal caregiver, 9059)

While the relationship can be a source of support, it can also lead to problems in the caregiving trajectory thereby negatively impacting the balance, as another caregiver points out:

> But this way / always this fighting / or the voice computer / she had not consented to a voice computer / but then she gets so angry if you don't understand her, but she says 'no' to the voice computer. And that / it's always these fights that we had with each other. But I already understand. It's just a little bit my mother. She just doesn't accept things. She has never / would rather bury her head in the sand and hope that it will pass. (current female filial caregiver, 5450)

Also, the balance was majorly impacted by person-related and relationship-related changes due to the disease. Neurocognitive and affective changes did preoccupy the caregivers, even though the former were more intricate than the latter.

> He has laughed crookedly at the beginning at stupid jokes, it is part of the disease, "sorry", he now said, "but it is also part of the disease" and of course he cried too, tears came much faster. Then the last time it saw the Christmas tree or that or at all, the illness with a thousand farewells, it really is like that. (former female spousal caregiver, 6697)

These behavioural changes and conflicts therefore not only affect how the caregivers are able to maintain a balance in the amount of support they can provide but also how they are able to manage everyday life for pwALS and themselves and organize support that would allow them to have some semblance of a normal life as well as respite support.

Managing everyday life

Despite the extraordinary situation of dealing with a terminal diagnosis, participants wished to continue everyday life as normally as possible. However, the terminal diagnosis of ALS impacts life plans. As an example, in one case of young-onset ALS, a current caregiver described the decision they faced, as they wanted to start a family.

> Then the diagnosis came and then we discussed how should it go on. And then we decided we would do a few months unpaid and go traveling and then soon afterwards the wedding, or we also went on a long honeymoon. And a child was still in question: yes / no, a child? We waited the first year to see what the course of the disease would look like. And it has a slow progression, so we figured ok let's try it. (current female spousal caregiver, 0493)

In this example, the normality of family planning is upset by ALS. Informal caregivers generally try to stick to a how their lives were before, consisting of everyday activities providing solace. Generally, family life was reported as being an important aspect of normalcy, next to work life. In the case that informal caregivers had other dependents to care for besides the pwALS, this additionally can become a source of burden; however, caregivers report getting support through their families. For informal caregivers in employment or self-employment, work life is a major part of this balance act.

> At one point you just noticed that it wasn't / that it was no longer possible with full-time work and that's why I actually planned / wanted to take a break ... And

> that actually went very well / thank God. But with my sister, for example, things looked very different now. She had a lot of trouble being absent from work to keep the appointments and we used all vacation days. (former female filial caregiver, 6217)

To enable the access of support structures, informal caregivers organized personal finances and received welfare support through health insurance and other social insurances which demanded administrative work. The situation of ALS and its progression made these normally mundane everyday tasks complicated and dealing with administration entailed uncertainty and helplessness. Furthermore, different conditions to receive institutional support had immense effect on the caregivers' ability to carry out caregiving.

> I felt completely helpless. I submitted the application for the wheelchair to the old age insurance and then they wanted 7000 things from me, and I work in the administration area in the office, so it's not like that I'm completely unaware of paperwork, but I didn't understand the letters they wrote to me, so really, really bad. (Former female filial caregiver, 8807)

Another aspect of upholding normalcy was keeping up leisure activities. Listening to music, singing in the choir, living a religious life, spending time with their dog are various activities informal caregivers undertake to balance out their lives.

Organising support

In addition to holding the balance and maintaining normality in their lives, informal caregivers are concerned with organizing support for pwALS to enable informal caregiving at home for the longest time possible. This support entails managing visits by friends or seeking social integration of pwALS. The informal support from other friends and family range from assistance of daily living, e.g. cooking and preparing meals, to hands-on nursing care. Getting or organizing informal social support and building networks were key activities of informal caregivers; these were helped if there were already existing peer-groups.

> Every Tuesday when we were at the (rehabilitation centre) we met (name of other ALS patient and caregiver), who still, he still has it, for 12 years. He was accompanied by his wife (…) Then she said let us meet half an hour earlier, than we can talk a little bit. (former female spousal caregiver, 6697)

Additionally, there are various sources of formal support that must be arranged as well. This includes, as for example, assistant caregivers, support and advice from experienced nurses specialized in ALS, and formal support through ALS centres. Informal caregivers need and value experienced and competent care personal. Generally, caregivers remarked needing a large multidisciplinary team to make care feasible at home, constituting of several professionals, sometimes privately paid. Ideally, this team would continuously provide support through the disease course to bereavement. Generally, caregivers appreciated if support was organised for them instead of having to organize it for themselves.

> Well, yes, from our situation it would have been best if the neurologist who made the diagnosis would have given my address to the care team and they had contacted me. (former female filial caregiver, 8807)

Accessing psychological and spiritual supports played a minor in our sample; however, where they were accessed, the care was important in enabling informal caregivers to deal with difficult feelings.

Searching for information to care for the pwALS is a crucial task that informal caregivers performed. Here, formal caregivers' knowledge and ability to provide relevant information as well as the ability of informal caregivers to gather information from other sources is crucial to adapt their care. The type of information extends beyond care and management to the cause of ALS opening up an intricate balance between blame and responsibility for having brought on ALS.

Another raised issue was the relocation of the pwALS to a nursing home. If informal caregiving is not able to be done at home, or not wished for, pwALS locate to nursing home.

> I couldn't sleep anymore, it was just too much of a burden for myself. Spitex came, but it just happened anyway / the responsibility was mine. I was always afraid that I would let her fall or / and then / so she said from the beginning that she was going to the nursing home, that's why it was never the question / rather "when?", But that she would go, we did from the start / yes, so I would have liked her to have stayed with us, but I would have / I couldn't do it on my own. And only with Spitex, I wouldn't have made it. That was the problem. (former female filial caregiver, 5450)

Here, problems are encountered in nursing homes such as that the care provided is not adequate for pwALS or communication problems arise. Hence, relocation to the nursing homes not necessarily provides relieve for caregivers.

Being present

One key work described by the participants is taking care of the pwALS in such a way that he or she does not feel alone and is not alone that it would endanger him or her. Hence, family caregivers are constantly planning to ensure that they or someone else is present with the pwALS. As long as the pwALS can stay at home on their own, it is easier for the caregiver to keep the balance as they have to organize less respite support and ensure some level of normal life for the pwALS.

In the caregiving trajectory, time needed to attend to the pwALS increases with disease progression. It can become round the clock care and attention with increasing debilitation depending on how much the caregiver takes responsibility for caregiving tasks or whether tasks are delegated to healthcare professionals. Therefore, the extend of being present depends on the availability of respite care.

> So / I'm sometimes tempted to say that I could do this (snaps fingers) and someone is standing here. On Mondays she [pwALS] usually goes to therapy in [city in German-speaking Switzerland], to the hospital / that is the hospital at / near the train station. Actually, the same woman always comes to go there with her. If she doesn't, I'll go, or / or someone else could come [...] Her sister comes here so that she is not alone. (current female spousal caregiver interviewed with her husband, 4739)

Informal caregivers report that if accidents had happened, e.g. a pwALS had a fall, this increased their vigilance and anxiety. This led some to hire round the clock help, co-habitating with the pwALS, and having to renegotiate privacy as professional caregivers moved in. The need to be available does not stop when the person cared-for enters the nursing home, as a daughter caregiver lays out:

> And that would / would actually already address the care in the [nursing home], but somehow we also want to give her the feeling that we are there and / she is actually not alone on any evening and we don't want that either she that is. (current female filial caregiver, 3847)

Keeping up with the illness

The progression of ALS is crucial for caregiving. A quick disease progression is seen as extremely burdening, however, caregivers' understanding of quick differed. Informal caregivers always felt that they lagged behind in caring for the impairments of ALS, such as dealing with urinary problems, keeping the body weight stable, for example, by inserting a feeding tube.

Depending on the progression, pwALS and caregivers make plans for advance care, treatment, and end-of-life decisions, as well as every day and bereavement related tasks. In one interview where the pwALS was present, it was revealed that while the husband had planned ahead for his own funeral, they were yet to decide on the place of death:

> Aha, um I talked to my husband. I don't know if he did something, but yes, we want to do that / we would do that / we would do that. Um, we discussed whether he would stay at home or go to the hospital or somewhere. Um but that's not yet / not yet decided. (female spousal caregiver, 6009)

Other plans centre around handling prognosis and managing expectations. Informal caregivers manage crisis events and difficult feelings like guilt for not being able to do enough. With the progression of ALS towards later disease stages, there is an increase in caregiver activity as they try to keep the pwALS mobile and involved in social life. Together with being present to the pwALS, this keeps them connected to the pwALS but at the same time, limits their social life and support. Thus, the end-of-life phase is characterized by shifting the balance of burden towards the family caregiver. Choosing the hospital with a palliative care station as a place of death can be a relief for informal caregivers, and they value hospitals providing such services:

> Yes, they were really amazing, they were really very good. My mother felt very comfortable just in the hospital we had every now and then / just / we / we had to go over at night because she was a bit panic - which has nothing to do with the nurses, but it was just the situation / she was better off in this palliative department because they knew better how, where, what. (former female filial caregiver, 6217)

The final phase of life of the pwALS is a critical time for family caregivers. At the end-of-life, coping and acceptance of ALS by the person affected is crucial for caregivers hold the balance in caregiving. Many problems arise if they do not, as one caregiver points out:

> Nevertheless, they could not offer this care in the way he wanted them to. He needs everything because he wants to get well again. Therefore, we had this discussion again this morning. He still does not see himself as a palliative patient, despite this extreme progression. (current female friend caregiver, 8694)

If the pwALS does not accept the terminal diagnosis of ALS, this can lead to conflicts with the informal caregiver as well as with the professionals. Furthermore, if they lose capacity to make decisions due to dementia or inability to communicate, family caregivers find themselves in moral dilemmas and conflicts with other family members. Hence, staying in communication with the pwALS is a significant issue. Together with the pwALS, caregivers accessed several technologies to enable communication at the end-of-life; however, even the most advanced technologies had problems.

> You also noticed because my mother couldn't speak anymore and things didn't always go well with this computer, they really came into the room and have already read through everything and said: "I've read this and that" and then my mother just had to blink. We explained it a bit like this and that and that actually works a lot through the eyes we also made a board with the most important things that you can ask if it is that could be. (former female filial caregiver, 6217)

Not only communication technology, but also ventilation masks and other devices are important from the perspective of informal caregivers during the final stages.

> But it worked, from there on we had to have an oxygen device, first at night, afterwards we had to have the oxygen device with the bottle and the backpack. (former female spousal caregiver, 0735)

In case informal caregivers received support and counselling from palliative care professionals on how to handle end-of-life situations at home, this needs to be head on:

> I found that actually / just good / you are talking about the same thing / there is plain text, so there is no beating around the bush. (current female spousal caregiver, 4739)

There where informal caregivers who reported not being able to support the pwALS at home for the final phase and having to relocate the pwALS to a care home or a hospital. However, it was clear that most caregivers really pondered the question how to provide care until death for the pwALS.

> Or how do you enable the family to look after someone at home in a good way? That is the reverse question for me / for a nursing home or an acute hospital one could have gone at any time. So you could have said at any time: call an ambulance and move [name of patient] to the hospital, but that wasn't the idea. We all wanted that, that she can be at home here. But how to set up the network at home for this care? (former male spousal caregiver, 1139).

At home, the responsibilities of organizing and administering medication fall onto the caregivers and their networks. Specialist's advice and care are needed that informal caregivers felt prepared to provide end-of-life care, even though some caregivers also pointed out that complete preparation was never possible. Especially the administration of morphine on part of the informal caregiver is a difficult activity. To enable the pwALS to die at home therefore means becoming more highly specialized in caregiving and providing palliative care in the home care setting.

> And then there is no rest either, I then injected morphine regularly, every 2-3 hours for weeks. And that was, yes, very stressful. (former female spousal caregiver, 3719)

At home, one important mediator for informal caregivers is the availability of respite and day support with the progression of ALS. In few cases, informal caregivers also used hospice care as a form of respite support. Generally, caregivers appreciated hospices as places where dying was not a taboo:

> It was also a happy house, there was no deathly silence in there now, not at all, you kept hearing people laughing. (former female spousal caregiver, 6697)

After death of the pwALS, not only bereavement and administrative related tasks were carried out by informal caregivers, but also searching for and accessing help with difficult feelings of grief, if needed.

Discussion

Our theoretical understanding underlined five categories. The core category, holding the balance in ensuring care and normalcy as disease progresses and until the end-of-life, is the social process of informal caregiving from the perspective of informal caregivers and therefore guides decisions surrounding caregiving. On this understanding, those caregivers that succeed in holding the balance are able to provide care at home until death. Holding the balance is heavily influenced by contextual factors of caregiving, e.g., relating to personal characteristics of the caregiver (*holding the balance, managing everyday life*), or activities of caregiving *(being present, organizing support, keeping up with the illness*) where the goal is to ensure the quality of life of the pwALS. As there is a heterogeneity of speed and subtype of progression of ALS, our work accounts for multiple caregiving trajectories – those closer to the slow progression of chronic neurodegenerative illnesses and cancer (Murray et al., 2005) and those closer to the unexpected death trajectory in acute brain injury (Creutzfeldt, Longstreth, & Holloway, 2015).

A similar study by Penrod and colleagues (2011) describes informal caregiving in their model of expected, mixed, or unexpected end-of-life trajectories as phases of *Sensing a Disruption, Challenging Normal, Building a New Normal,* and *Reinventing a Normal*. In the context of the expected death trajectory such as ALS, reinventing a normal is replaced by *Losing a Normal* (Penrod et al., 2011). These phases help those experiencing the situation make sense of adaption to ALS but are too unspecific. While our understanding of caregiving is compatible with “phases [that] are demarcated by key transitions experienced when the illness progression manifestly challenges the established steady state achieved by the caregiver” (Penrod, 2012, p. 174), in ALS this progression is foreseeable. Hence, the work informal caregivers do is not only reactive (achieving the steady state after crisis) but also anticipative (upholding the steady state). The basic process of holding a balance as outlined in our study is not directly connected to an understanding of normality. This is because we do not want to ascribe normality to the difficult process of holding the balance, even though rendering this into terms of “new normal” might be fitting. We emphasise upholding normality in our model in the category of managing everyday life. Nonetheless, managing everyday life is also a source of burden and work.

Much of the support provision is mediated by the caregiving relationship. Bassola, Cilluffo, and Lusignani (2020) used Grounded Theory to shed light on interpersonal processes involved in the caregiving dyad in ALS. Their analysis yielded three main categories "'reciprocity', 'loving to care' and 'changing to care'" and four secondary categories: 'having support', 'sharing suffering', 'protecting each other' and 'thinking positive'" (p. 3). While interpersonal processes were not the focus of our analysis, these categories outlined here map onto our findings that caregiving depends on intra- and interpersonal processes (e.g., acceptance of diagnosis on part of pwALS).

Our theoretical understanding is flexible in explaining caregiving and decisions of informal caregivers and their families. It underlines the importance of stressors (bringing the caregiver and family system out of balance) and resources (activities which help the caregiver) that shift the balance in favour or against informal caregivers. This flexibility is needed for any theory of informal caregiving in ALS, as disease progression, personal characteristics, welfare state support and dynamics within the family systems differ in every case.

Limitations

This theory's flexibility is also a limiting factor; the theoretical understanding of informal caregiving in ALS has its utility in being adapted to individual cases and as a heuristic to approach informal caregiving in ALS for healthcare providers, family members, and others. The key approach resulting from our theoretical questions is resource-oriented but also looks at needs for support in individual cases, *what* exactly might help informal caregivers to hold the balance. Furthermore, this theory is only broadly testable because it is circular; if one would find that the balance had shifted negatively, necessarily the caregiver would experience burden or distress. Therefore, our theoretical understanding primarily adds to the existing literature on caregiving processes in ALS. Together with these other studies, it can be used to paint a rich picture of informal caregiving in ALS, especially during the later caregiving stages.

Conclusion

This study suggests that in a constant endeavour to hold the balance, informal caregivers for pwALS provide care and render decisions. This theoretical understanding helps if informal caregivers lose their balance in the tightrope-walk of providing care to a dying person and interventions and support by healthcare professionals can be directed to shift the balance in favour of the caregivers.

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Chapter 6.

Bereavement and support experiences of informal caregivers of persons with amyotrophic lateral sclerosis: A qualitative study

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Bereavement and support experiences of informal caregivers of persons with amyotrophic lateral sclerosis: A qualitative study

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Abstract

Informal caregivers provide the major part of care for persons with amyotrophic lateral sclerosis (ALS), a terminal neurodegenerative disease. Apart from providing care, informal caregivers are themselves in need of support to fulfil the task of daily caregiving and to reduce their burden of caregiving. This need for support does not end with the death of the person cared for. In this study, we explore the themes of bereavement and support experience of informal caregivers of persons with ALS from interviews conducted with bereaved informal caregivers (n = 14) in Switzerland. Three key themes were salient in our data: *Reacting to bereavement, finding support*, and *adjusting to life without the person with ALS.* These themes are contrasted with themes from interviews (n = 11) with healthcare professionals (nurses, therapists, physicians) who care for patients and families with ALS. Themes described were *offering support* and *identifying gaps in the support*. We discuss support after bereavement for informal caregivers of persons with ALS in the established public health model of bereavement support. Bereavement support needs to be proactive from healthcare professionals; however, it requires goodness of fit to address those in need and not those who are adequately supported by informal sources of support.

Keywords: informal caregiving, amyotrophic lateral sclerosis, bereavement, needs, qualitative study, family

Word count: 5450

Bereavement and support experiences of informal caregivers of persons with amyotrophic lateral sclerosis: A qualitative study

Amyotrophic lateral sclerosis (ALS) is a terminal neurodegenerative disease that leads to progressive muscle dystrophy. Given the inexorable nature of ALS, any care provided to people with ALS, by formal or informal caregivers, can be understood as palliative care. From the onset of the disease, the palliative approach in ALS therefore includes the communication of diagnosis, assessment and management of symptoms like saliva management, interventions such as gastrostomy and ventilatory support, and end-of-life care (Andersen et al., 2012, Oliver & Aoun, 2013; Foley, Timonen & Hardiman, 2014; Oliver, Borasio & Walsh, 2014; Goutman & Simmons, 2018; Oliver, 2019). It is generally understood that supportive care extends “beyond the period of care for the patient, and includes a consideration of the need to support and counsel those who have been bereaved” (Sepúlveda et al., 2002, p. 92).

Informal caregivers, mainly family members, provide supportive and palliative care throughout the ALS disease course. At home, they address symptoms of ALS such as fatigue, immobility, dysarthria, pain, difficulty sleeping, dysphagia, dyspnea, weight loss, mood disturbances, depression as well as neurocognitive and behavioral changes, and are responsible for medication management (Lerum et al., 2016; Brizzi et al., 2020). Patients and informal caregivers are supported by professional multidisciplinary care which has been implemented in many countries since the turn of the century (Sukochienė et al., 2020). As part of caregiving, informal caregivers experience different supportive needs based on the level of care given and disease progression (Galvin et al., 2018). Due to bereavement, informal caregivers experience distress and mental health disorders such as prolonged grief disorder, anxiety or depression (O'Brien et al., 2016; Aoun et al., 2012). In addition, informal caregivers with poor family functioning, spousal relationship with the deceased person, and insufficient support before bereavement are at risk of prolonged grief disorder (Aoun et al., 2012). Following Zisook & Shear (2009) in this paper the term bereavement is used to refer to the fact of having lost someone to death, while the term grief is used to describe the emotional, cognitive, functional and behavioral responses to such a loss.

Most studies have focused only on psychological, emotional, existential, and social sequelae of bereavement of family caregivers of people with ALS. Little is known about the full spectrum of bereavement needs of informal caregivers and their experience of support (Poppe et al., 2020). For support of informal caregivers, Hebert and colleagues (2004) reported that although healthcare providers think grief and bereavement support for caregivers is important, their ALS care centers often did not provide adequate support that extends beyond the death of the person with ALS. They concluded "studies are needed to obtain ALS caregivers' opinions about the grief support offered" (Hebert et al., 2004, p. 138). While some studies explored the bereavement phase as part of a wider study on informal caregivers' end-of-life caregiving needs (Whitehead et al., 2012), a few have directed their study aim at the bereavement phase. An exception is the study by Martin and Turnbull (2002) who have surveyed 32 families of people who have died of ALS. They found that "ALS caused lasting emotional and financial hardship in families, and 37% of respondents felt they were coping poorly, sometimes years after the patient's death" (p. 181). A recent survey from Australia found that 40% of ALS informal caregivers felt that supportive needs were not met after bereavement and that support by family and friends was most helpful (Aoun et al., 2020a). The authors concluded that there is a “need for a new and enhanced approach to MND [motor neurone disease] bereavement care involving a caregiver risk and needs assessment as a basis for a tailored ‘goodness of fit’ support plan” (Aoun et al., 2020a, p. 1). In this article, we want complement these survey studies with our qualitative data. Thus, the goal of this paper is to highlight experiences of bereavement support reported by two stakeholder groups: family caregivers of people with MND/ALS and healthcare professionals providing care for individuals with ALS and their families.

Methods

Sample

Our overall sample comprised of 24 informal caregivers, mostly family caregivers, with an age range from 29 to 73. From the family caregivers, most (96 %) were spouses or daughters of individuals diagnosed with ALS. Ten informal caregivers were currently providing care to a person with ALS and were therefore excluded from analysis on bereavement experience and needs. Furthermore, we conducted 11 interviews with healthcare professionals (physicians, physiotherapists, nurses) providing care to individuals with ALS and their families. Informal caregivers were recruited purposefully through ALS centers in German speaking part of Switzerland, through patient initiatives (Verein ALS Schweiz), peer contacts of former caregivers, and snowball sampling. For the purpose of this article, only interviews of former informal caregivers after at least one year from bereavement were included. Therefore, the dataset for the analysis was made up 25 interviews (n=14 bereaved informal caregivers, n=11 healthcare professionals).

Participants contacted the researchers via phone, mail or email after having heard of the study. With caregivers, interviews took place mainly in person in the participants' home to accommodate time and caregiving constraints. Healthcare professionals were mostly interviewed via telephone. Interviews were conducted and audiotaped after all participants had provided written informed consent. Interviews were between 30 – 130 minutes in length. Each participant was interviewed only one time during the time of 2018-2020. Interviews were transcribed verbatim in Standard High German using MAXQDA (Verbi Software, 2020).

The interviewers were a female physician (MD) working in general practice and a male clinical psychologist (MSc.) working in the academic setting. Both were native Swiss German and Standard High German speakers. Both had prior practice in conducting qualitative research interviews or analysis and were repeatedly trained in qualitative data collection. The first four interviews served as a pilot for the semi-structured interview guide and the interviewers' interview skills, which were supervised by a senior researcher. For informal caregivers, the interview guide comprised of questions regarding practical supportive needs during caregiving, difficult situations, advance care planning and end-of-life, as well as the time after bereavement. For healthcare professionals, interview guide centered on support for caregivers, also after bereavement. The interviewers did not have any prior relationship to the research participants, nor did the participants have specific knowledge about the interviewers apart from general information about the project.

Analysis

We conducted an inductive thematic analysis within a critical realist framework – meaning that we viewed the data as an assumed reality of informal caregivers' experience of bereavement (Braun & Clarke, 2006). Correspondingly, we approached this reality by inductively coding segment by segment of the interview data. Segments consisted of units of meaning (usually one to three sentences). This meticulous coding upholds rigor of our analysis.

Data analysis was performed in parallel to data collection. Data saturation was monitored while inductively coding and achieved when recurrent coding occurred. Data saturation was therefore understood as post-hoc information redundancy achieved during data analysis (Braun & Clarke, 2019). Initially, eight interview transcripts were coded by hand by teams of two to three coders (six coders overall) to satisfy inter-coder agreement of approach to the data. Later, interviews were coded inductively using MaxQDA solely by the first author. Inductive codes were grouped by thematic similarity making up subthemes. Several subthemes formed themes. In the results section, we present the results from the analysis of interview of informal caregivers and healthcare professionals in two parts to provide a comparison of themes in the discussion.

Ethics approval

The project was submitted for review by the cantonal research ethics committee (Ethikkommission Nordwest- und Zentralschweiz, EKNZ) through swissethics. As the project does not qualify as research with human subjects under Swiss laws, a certificate of non-objection was issued by the EKNZ. Despite not being subject to human research laws, we felt that it was necessary to uphold the standards required by the same laws of Switzerland, that is, having written informed consent of every participant, as well as storing data separately in a de-identified manner. Furthermore, no identifying information is presented in the paper that could jeopardize confidentiality.

Results

Bereavement experience of informal caregivers

For informal caregivers, the thematic analysis resulted in three separate themes: reacting to bereavement, finding support, and adjusting to a life without the loved one.

Reacting to bereavement

Several caregivers expressed that they felt severe burden or grief after bereavement. They noted how bereavement after death from ALS was different due to the extensive nature of caregiving provided to the person with ALS. A widower who had cared for his wife with ALS at home succinctly described the bereavement as different to other deaths in light of the exhausting and long caregiving needed for his wife.

> It is not like that / I have already experienced several deaths: parents, brothers and everything else … That was much less burdening, but if you have it in a way where you are still exhausted, where the reserves decrease and then comes the case X. … [ehm] I was amazed how much energy you get when you need it. Because I don't know where you get it from. And afterwards, especially when you have a family with children. To put it poorly, when the coffin is gone, life goes on, right? And you have to be there again. (65-year-old husband, 1139)

Moreover, almost all informal caregivers reported feelings of stress and depression, self-blame, and being stuck in grief. They stated that it was difficult to live normally, that is, they were unable to undertake activities or (return) to work. Sometimes, informal caregivers got ill after bereavement. Grief in some cases only manifested later as the caregivers were pre-occupied with burial or other tasks after immediate bereavement.

> I will/ I have now a little bit the feeling - it has been one year now and I have now a little bit the feeling that the second year is almost worse than the first one. It's like that/ it's like it's fin/ it's all finished, you don't have to do anything anymore. (73-year-old wife, 0735)

As caregiving had ended, informal caregivers reported working more, rushing into life, visiting friends or not being able to do something with the newfound time. One caregiver reported actively keeping busy against the estranging effects of being alone after bereavement:

> P: When you are alone, you become a little special, so [I mean] strange sometimes. Now I go to a university group [...]. And then we sometimes deal with such topics [end of life] again and then you are simply involved, whether you want to or not, that's still good. And sometimes [I go] to the education center, a lot to the cinema. Yes, I do a lot with my daughter.
>
> I: You are "busy" (laughs), really.
>
> P: Yes, yes I am. Yes, maybe sometimes too busy, that can also be. It can also be an escape again. Yes, yes, you can interpret it as you like. (72-year-old wife, 5192)

Some caregivers described bereavement as a natural process of moving on, where time was needed to deal with grief. They noted the absence of the late person and grieved its finality. Furthermore, they reported taking time off work or having already been prepared to deal with the death of the person with ALS. Generally, pointing to the normality of grief for most caregivers, one daughter of a person with ALS described not needing to access psychological support due to the non-traumatic nature of death from ALS in her case:

> Neither of us did. But I think simply because it / it was somehow good and it wasn't a traumatic experience for us actually / it was bad, sure, but it wasn't [ehm] / I imagined it worse. (Young adult daughter, 9352)

Some caregivers described happiness because others had said they had done a good job, for example healthcare professionals. For one caregiver, recognition for the work she had done came also from a formal carer providing round the clock assistance:

> She wrote me later in the letter that she was actually always astonished how my husband accepted it. And for her, we were like role models, especially me, how to deal with a sick person. I was a role model for her. (73-year-old wife, 0735)

If support was wanted after bereavement, informal caregivers described several sources where they found it.

Finding support

Informal caregivers reported that they searched for support themselves, showing that information about where and how support was available was not provided to them. Those who found support revealed that these were available from different sources. For professional support, informal caregivers accessed psychological, medical and spiritual care. Psychological support was found in most cases in psychotherapy sessions or, if rare, inpatient psychosomatic care, which only one caregiver described.

> And it went more downhill, I was so stressed, felt so weak that I ended up just lying in bed, they said that is depression, I said I am depressed because I cannot

> do anything anymore. So I saw it the other way but until I was so done after two and a half months I said to the family doctor "you have to register me in a clinic" and then I was in [city name) in the psychosomatic clinic, 6 weeks and that pulled me out of the swamp. But I am still not, it keeps coming back, I still can't live normally. (73-year-old wife, 6697)

Informal caregivers noted the importance of having continued support starting prior to the death of the person with ALS. In one case, a psychologist provided continued support for the family throughout the disease course. After death, she acted as an intermediary handing farewell letters from the person with ALS over to the bereaved caregivers:

> Yes I am [ehm] / my mother went to a psychologist when she got sick and I went to her [psychologist] once [ehm] at that time and [ehm] now after that I was there again, exactly. [...] And it was also helpful because she knew my mother and actually accompanied us during the illness / well yes for a little bit. ... Yes and my mother also gave her letters [ehm], which were supposed to be handed down to us us. And that's why we were actually well looked after by the psychologist and my sister recently went to her again. (28-year-old daughter, 6217)

Also for medical support, some participants highlighted that they stayed in contact with the medical professionals who had cared for them and the person with ALS.

> So (pause) finally I have to say [hm] of all support / the support from back then [before bereavement]/ the nurse is now a friend of mine, [hm], she worked at Spitex [home nursing care services], and was there for me. I think that [she] really became a very close contact. (65-year-old wife, 0751)

Correspondingly, the same participant remarked missing continued support by healthcare professionals after bereavement:

> I’ve seen the others [professionals] once but not after that. And haven’t seen them since. No one asked: “How are you doing” Or something like that. Nothing came from that place. (65-year-old wife, 0751)

Apart from professional support, informal caregivers found support in the form of peer-support, i.e. grief cafes offered by the church or self-help bereavement groups.

Informal caregivers missed support relating to administrative work. This related to lasting power of attorney for accessing bank accounts. If these documents were not filled out with the signature of the late person with ALS, accessing bank accounts could become a matter of impossibility.

> So one must actually in time, as long as people with ALS are still able to authorize, or even earlier, also with the banks on the bank form, the powers of attorney. We do have money, but they won't give it to me, will they? Even if you / just / I am / I have done this with a colleague, notarized every document. Certain banks have [that] / not that long [we did that] / you just have to do, otherwise there will be problems with the banks. Since I had so much to do with the business and with them, it worked out fine. At the end I brought them the federal court decision and said: "Tell that to your legal service in (City), that's not how it works, it has to stop”. Yes, if someone doesn't know that now and has one or two accounts at the bank and then the man dies, now as an example, and then the woman is there or vice versa and then / can't get at the money. (65-year-old husband, 1139)

As this caregiver points out, due to the progressive of nature of ALS, these powers of attorney often had to be filled out already in quite early stages when the person with ALS was still able to sign the documents. Another issue brought up by a few caregivers was filling out tax returns. One caregiver complained

> What annoyed me most, that is administration. So, the woman is dead and you have to fill out the tax return within 30 days. [...] So... And that is actually not the most important thing in this situation, but that is disturbing. (70-year-old husband, 4673)

This goes hand in hand with the quoted caregiver above who described this as "you have to be there again" (65-year-old husband, 1139), hinting at the ordinariness of administrative tasks amidst severe grief.

In general, all caregivers described having received support from their social network, from family and friends or at work. They valued friendship and having contacts in the neighborhood. They described active endeavors to gain contact to others. Most caregivers expressed that this was due to the need to talk to someone and not being alone:

> And I'm a very communicative person, I just go out, I do it also now, I go running every day, you'll meet somebody again somewhere and/or I have a great girlfriend who came almost weekly during the last half year when my husband was ill. She also comes a lot now. It's easy, then we can have a quick coffee here and talk for an hour or two together, that you just TALK about it. (74-year-old wife, 0735)

However, one daughter also noted how it is difficult to talk about grief if the other person was unaware of the exceptional situation of ALS before death. A few informal caregivers described having rituals to cope with grief. They reported having dialogues with the dead or visiting the cemetery. One caregiver expressed:

> Yes, I have already, I went there every x times to the cemetery and everything. I still go there every month. It's terrible, I still talk to him. (72-year-old wife, 5192)

However, some participants did not see the need for support after bereavement at all:

> Not, actually not [having need for psychological support]. I think though, I could have gone maybe to my general physician. Pretty sure I could have. And with him / I mean we knew each other well. (57-year-old wife, 3719)

While these were the sources of support informal caregivers accessed or did not access, generally life after bereavement changed, and informal caregivers adjusted to it.

Adjusting to life without the person with ALS

After the death of the person with ALS, some spousal caregivers had new partners. Those who had new partners revealed that it resulted in challenges. For example, their children were not accepting the new partner.

> Well, that's a bit difficult, so I have my life partner - he did the faith course with me. Um, we got together soon … Because he lost his wife to cancer the same year as I lost my husband, we could talk about a lot of things. The difficult thing was just to accept the new one for the children… (65-year-old wife, 6598)

In general, family caregivers mentioned support needs for bereaved children as “the children themselves grieved, each in their own way” (72-year-old wife, 5192). A few participants also stated that support for children came mostly from the family or from schools. These family caregivers mentioned that psychological support for children would have been helpful but was not available. Participating caregivers also mentioned psychological growth on their and their children's part.

> Our son who helped me care for her – I am still astonished today. He really did that. He took a big leap in dealing with social life at that time, which we still notice today. It is incredible. (65-year-old husband, 1139)

This caregiver also compared this to other terminal illnesses, where they expected similar personal growth.

> But [name son] was at home all week. He really, yes, he noticed the decay of his mother every day. That's quite formative, you're right. I don't think that's anywhere, it's as good as cancer, you would have to take that into account just as much in the case of cancer. Cancer and ALS and all these diseases. (65-year-old husband, 1139)

The adjustments to the life without the person with ALS highlighted not directly support-related needs. The next section delineates support offered by healthcare professionals.

Bereavement support by healthcare professionals

For healthcare professionals, the thematic analysis resulted in two separate themes: offering support and identifying gaps in the support.

Offering support

Healthcare professionals delineated forms of institutional support they usually practiced after the death of a person with ALS. Practical support included writing a bereavement letter to the family, having a church service next door to the hospice, and gathering remaining care aids (beds, wheelchairs, non-invasive ventilation masks) from the homes of the families. One justification healthcare professionals relayed for these home visits is to make life easier for informal caregivers:

> And afterwards I only make home visits when the patient has died and had a ventilator. [ehm] Then I go to the families so they don't have to return the ventilator to the hospital, that's important to me. It is always almost like that, a little bit humiliating, the patient has died, you pack your things and have to go back to the big house. (female nurse working as a care manager, 987)

More than providing direct support as in home visits, healthcare professionals offered or referenced possible means of support. Most of the healthcare professionals offer one-time talks, telephone calls, or other contact after bereavement. Furthermore, healthcare professionals report referring informal caregivers to psychotherapy or group support.

Identifying gaps in the support

While group support was frequently referenced, there were also concerns regarding accessibility of bereavement support. They highlighted that informal caregivers would have to be proactive to access support. In addition, most healthcare professionals wanted to provide more support to the informal caregivers after bereavement:

> The relatives still need to know: what support there is and who is fit, who has time, who has a good school education, who speaks the language here, who is culturally rooted here, they can access support, and everyone else can't. And that is ethically very off-putting because it is not fair, huh, if we had money, personnel, time, resources, then we would love to offer such an afternoon table, we would love to offer courses for relatives, modular, on different topics, and also separated by diagnosis ... (female palliative care physician,167)

In line with the concerns about accessibility were the agreement of most healthcare professionals that many caregivers did not access support. According to the healthcare professionals, this was because informal caregivers may have other means of support or that it depends on the relationship with them.

> So it is always said that palliative care includes bereavement care, we also have a grief coffee group, that is right and we also have a follow-up care by our psychologist, if that is desired. Anyway that the relatives and the doctors can come again to discuss their thoughts again or, you know. I think it is not used as I would expect it but as I said that is felt truth, I cannot give you a number. (male palliative care physician, 810)

One palliative care physician pointed to the fact the structure of support relies on the caregiver’s initiative – which might not be what is needed after death. Healthcare professionals said that having bereavement care serves both caregivers as well as the involved healthcare professionals as a form of psychological closure. Healthcare professionals were also worried, like our family caregivers, that children were not adequately supported if a family member died of ALS.

> So I know two examples. I don't know what the need would be for the young people... I don't see enough in this area, but I now have some families where the father will die one day, and what happens afterwards, how do the young people deal with it, you would have to ask the relatives who have already lost someone, or families who have already lost someone, that makes me wonder. (female nurse specializing in ALS, 874)

Discussion

In reaction to bereavement, informal caregivers report several forms of psychological strain and needs. The reported psychological sequelae range from describing the normality of grief with coming to terms with the absence of the late person to expressing severe distress and the need for mental health bereavement support. This is similar to the results of the study by Whitehead and colleagues (2012) where informal caregivers "experienced complex emotions and distress" during the period of bereavement.

Whitehead and colleagues (2012) also found that administrative processes put a strain on informal caregivers to which the results of this study add different sources of administrative burden (e.g. tax, access to bank accounts). Bereavement bureaucracy might be an understudied field of caregiver burden, as the field has mainly focused on psychological, social and emotional sequelae of bereavement. However, other authors have argued that informal caregivers can find meaning after bereavement in bureaucracy (Wadey, 2013). Nonetheless, this presupposes, the author argues, that the sense of bereavement bureaucracy is clearly communicated (Wadey, 2013). Indeed, in our study, informal caregivers report about the pointlessness of bureaucracy. This remains an understudied but important aspect of bereavement research in ALS and in palliative care in general.

The present study also highlights changes to family life, with children being in need of support and challenges posed by new partners. On a practical side, there is the need to sell or return or give away care aids families had received or bought for the persons with ALS. These, now unused, mobility and care aids add to the financial needs already established by the survey of Martin and Turnbull (2001).

There are different lines of support for informal caregivers after bereavement. Aoun and colleagues (2012) delineated three pyramidical components relating to informal caregivers' level of need of bereavement care from the public health approach. The three components are first that "all the bereaved people should have access to information about bereavement and relevant available supports», second, that they should have "formal opportunities to consider their loss", and third that they "need specialist intervention such as counselling, mental health services, bereavement services, or psychotherapy" (p. 15). Each component is more specialized than the one before; the first relying mostly on informal social support and on physicians, social workers and psychologists involved in primary care, the second more formal opportunities such as grief cafes in this study, and the third dedicated psychotherapy or other specialist support offers. As the experience of informal caregivers who participated in our study maps onto this approach, we will discuss informal support and formal bereavement support distinctly.

Informal support by friends and family

This study supports the idea that most of the first component of the public health approach as outlined above can be covered by informal support, that is, friends and family. Healthcare professionals in our study pointed to the fact that informal support might substitute support they offered through their institutions. All caregivers highlighted wanting someone to talk to and being attentive towards their possible needs. Most informal caregivers expressed that the latter softened the blow of bereavement. Family and friends are reported to be most important, even though caregivers also pointed to the limits friends can have as confidantes if they did not know the situation before death from ALS well. That informal support is crucial has been established in bereavement research (Vachon & Stylianos, 1988) but might be generally helpful in all distressing situations, not specific to bereavement (Stroebe et al., 2005). Informal social support can help to cope with loneliness or fear of loneliness, as illustrated by our results. Friendship and community are highlighted as important by informal caregivers. For the second component, informal support can also extend to organized peer support in grief cafes as well as everyday group activities such as choir singing.

Formal bereavement support: two sides of one coin

In their view of formal support, both participant groups showed wide variation. Healthcare professionals expressed the wish to offer more support, while informal caregivers were often content with the support offered, but reported having to search for the support themselves. That initiative had to be on the side of the caregivers was reported overwhelmingly by healthcare professionals as well. For informal caregivers, passive access to support where they have to take the initiative might not be as essential as proactive support by healthcare professionals. Nonetheless, this offers a fine line to walk. Offering too much bereavement support by healthcare professionals risks iatrogenic harm of medicalization of grief. Contrarily, offering only passive support runs the risk that caregivers who might be in need of support do not access it. The “goodness of fit” (Aoun et al., 2020a) between bereavement support and needs is thus not only of utmost importance, but might possible be also established beforehand when informal caregivers are informed about possible support options after bereavement. Informal caregivers and healthcare professionals highlighted the role of (young) children as especially vulnerable groups for bereavement care. Bereavement care for this group is understudied, both in the context of ALS and in general.

Bereavement care from a public health and palliative care perspective

A general challenge for bereavement care in ALS is that informal caregivers in ALS tend to focus more on the needs of the person with ALS than their own needs (Galvin et al., 2018). Hence, as there is a lack of death preparedness on part of bereaved informal caregivers (Mc Veigh et al., 2019), there is a vital need for the integration of death and bereavement preparation within integrated neurological and palliative care for ALS (Oliver, 2019; Sukockienė et al., 2021). Establishing coordinated care by multidisciplinary teams and specialist palliative care services from diagnosis as recommended by guidelines (Andersen et al., 2012) is one of the prerequisites for integrating bereavement care into clinical routine. Generally, multidisciplinary care needs close collaboration between neurology and palliative care services, with increasing training of all involved (Oliver et al., 2016). Ideally, these specialist services should evaluate for the burden of grief and check-in with informal caregivers after bereavement. Further research should establish what pre-bereavement risk factors account for complicated grief in informal caregivers of people with ALS.

In contrast to this specialist bereavement care in ALS, a public health approach to bereavement support needs to be explicitly pragmatic and broad (Aoun et al., 2012). Overall, Whitelaw and Clark (2019) note the lacuna of interdisciplinary interaction between public health and palliative care without adequate integration of palliative care in wider health and social care systems (Cohen & Deliens, 2012). Public health approaches to bereavement, such as the Healthy End of Life Project (HELP, Grindrod & Rumbold, 2018), have centered around building community capacity to "support members of a community in caring for each other at the end of life" (Grindrod & Rumbold, 2018, p. 73; Rumbold & Aoun, 2013). In the last years, community capacity development as easy accessible social support for people at the end-of-life and their families has taken the form of death doulas (Rawling et al., 2019), compassionate communities (Breen et al., 2020; Abel, 2018), or hospice volunteers (Morris et al., 2015). Integrating bereavement care within these forms of support can benefit informal caregivers who might not need specialist attention after bereavement but are in need of solace and comfort. Therefore, bereavement care for informal caregivers of people with ALS needs both attention of palliative care specialists as well as recognizing that loss and bereavement go beyond individualistic incidents and touch upon shared human experiences within communities.

Limitations

This exploratory study provides a qualitative account of bereavement experiences and needs in informal caregivers of people with ALS in Switzerland. In this way, it is tentative evidence for specific bereavement needs of informal caregivers; however, it is not generalizable. For example, the needs relating to administration are specific to the Swiss context. It is likely that needs differ depending on the administrative context. Furthermore, our sample of informal caregivers was self-selected as they approached the interviewers for participation in the study.

Conclusion

Informal caregivers experience several bereavement needs after expected death of a family member or friend with ALS. Apart from psychological distress and grief, bereavement bureaucracy is reported as burdening. Accessibility and proactive support by informal and formal sources are reported as crucial. Bereavement care in ALS should be provided on the basis of goodness of fit and established before bereavement. The effect of bereavement on children who might have been young carers to persons with ALS seems to be a neglected field of study.

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Chapter 7.

Brain-computer-interfaces, completely locked-in state, and end-of-life decisions

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Brain-computer-interfaces, completely locked-in state, and end-of-life decisions

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Abstract

In the future, policies surrounding end-of-life decisions will be faced with the question whether competent people in completely locked-in state should be enabled to end their life via brain-computer interfaces (BCI). This article raises ethical issues with BCI-mediated action in the context of end-of-life decisions, more specifically regarding withdrawal of life-sustaining ventilation and assisted dying. We argue that enabling patients to end their life even once they have entered completely locked-in state might, paradoxically, prolong and uphold their quality of life.

Keywords: brain-computer interfaces, assisted suicide, withdrawing treatment, end of life, ALS, locked-in state

Background

In the last decade, brain-computer interfaces (BCI) have been developed to the point that they are clinically useful (Holz et al., 2015). Given the rise of BCIs designed to assist people with brain injury or neurodegenerative diseases, a new kind of non-bodily physical action might become possible. Recently, Rainey and colleagues (2020) outlined moral responsibility for BCI-mediated action understood as "willed bodily movement". This kind of movement is the only movement available for people in a completely locked-in state (CLIS[1]) who lack all voluntary non-BCI-bodily movement, including eye movement. However, people in CLIS are not only metaphorically locked-in, they are also "through social exclusion, stigmatization, and frequently being underestimated in their

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[1] Locked-in state has traditionally been separated into incomplete, locked-in, and completely locked-in state.

abilities, […] locked out", as Johansson and colleagues (2017) have argued. In this article, we want to draw attention to restrictions of autonomy at the end-of-life of people in CLIS.

Notwithstanding multiple practical and technological problems, this paper highlights ethical issues with assisted dying policies based on self-administration-models in CLIS where patients can only control the lethal substance intake via BCI. Self-administration by the dying person is a fundamental requirement in most countries where assisted suicide is legal, including in Switzerland. That is, the person wanting to die has to have physical control over the pharmakon. Physician-assisted suicide (PAS) is legal in Switzerland if the assistance is not provided because of selfish motives according to the Swiss Penal Code, Article 115. Access to PAS requires decision-making capacity (henceforth, capacity) and physical control over the lethal drug. Physical control can go as far as having a mechanical device linked to muscle activity opening a gastric catheter containing pentobarbital (Bosshard, Jermini, Eisenhart, & Bär, 2003). This device renders it possible to "provide an easy-to-handle remote control which they can activate with a small movement (e.g. a finger, toe or jaw) to start the attached pump"[2]. In CLIS, this movement is impossible.

In self-administration policies, both criteria, capacity and control, serve to safeguard autonomy in these end-of life decisions. In this article, we argue that in some cases of CLIS, more specifically for people with CLIS in the context of amyotrophic lateral sclerosis (ALS), PAS through BCI should be an option in legal regimes allowing PAS under self-administration. To do this, we first provide reasons why it is important to discuss physician-assisted suicide in the context of CLIS at all, given that withdrawing life-support constitutes a more obvious way to end one's life for these patients.

Withdrawing treatment and physician assisted suicide

People in CLIS are ventilator-dependent and in need of artificial feeding. Hence, if they are legally competent, they do not need to commit PAS, as withdrawal of treatment (WT) offers them a viable way of ending their life. Unlike PAS, the latter is legal in the vast majority of countries. But also beyond legal considerations, switching off life supporting machines seems more reasonable than an invasive procedure that involves

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[2] Dignitas, „How Dignitas Works", Brochure, http://www.dignitas.ch/images/stories/pdf/so-funktioniert-dignitas-e.pdf (15.02.2021)

administering pentobarbital through a gastric catheter. Still, for the purpose of this article, we focus on PAS via BCI since we believe that our arguments concerning PAS could be similarly applied to WT. The most notable difference in PAS and WT is that PAS needs a wilful action (the person's physical control of the pharmakon), while WT solely needs the communication of treatment preference to the attending physicians (i.e. for the ending of treatment). In this way, PAS has more extensive requirements subject to ethical analysis. It is central to our argument that if these requirements can be met for PAS, they can also be met for WT.

In addition, some patients in CLIS might also have reasons to prefer PAS, e.g. because it gives them better control about the time of their death. Arguably, like other patients, patients in CLIS should have the choice between PAS and WT, where legal. This choice however is subject to having decision-making capacity and, in PAS, having physical control.

Decision-making capacity as a gatekeeper for PAS via BCI – the case of ALS

In most cases of CLIS, capacity is difficult to establish because of the lack of adequate and reliable communication. For example, Cabral and Illes (2017) argue that communication via functional magnetic resonance imaging fails to establish capacity in patients with traumatic brain injuries in general. In CLIS, as long as communication with patients via BCI is unreliable, any evaluation of decision-making capacity will be impossible. Therefore, it would appear that without urgently needed progress in the development of BCI communication, the use of BCI for end-of-life decisions remains inadequate, as an autonomous, capacious, decision cannot be reliably communicated.

However, there are more clear-cut cases of CLIS where capacity seems preserved. For example, there are people with amyotrophic lateral sclerosis (ALS), who remain cognitively unimpaired (Fuchino et al., 2008). ALS is a terminal neurodegenerative disease with progressive muscle weakness and atrophy leading to locked-in state, if patients are on invasive mechanical ventilation. While in locked-in state, eye movement is preserved, enabling communication by eye movement. However, some patients suffering from ALS unfortunately enter CLIS (Murguialday et al., 2011). For these albeit rare cases, BCI communication seems urgently needed but has not been successfully established to date. However, for most people with ALS who present with CLIS, it seems that capacity is often present as they are cognitively unimpaired and might be tested in the future through BCI-mediated language use. If BCI becomes sufficiently reliable, this would enable that capacity could be probed and evaluated.

This form of testing would be particularly relevant since some ALS patients do lose capacity due to cognitive decline caused by some forms of ALS, encompassing “personality change, irritability, obsessions, poor insight, and pervasive deficits on frontal executive tests” (Phukan, Pender, & Hardiman, 2007). If reliable communication can be established through BCIs, this would permit to distinguish between absence and presence of capacity for different types of decision-making, including end-of-life decisions. However, as long as there are narrow constraints on the complexity in communication via BCI, there will probably be a number of cases where it is impossible to establish whether a person has capacity to make an informed decision or not. Depending on whether BCI communication will allow more complex communication about the decision at hand in the future, capacity might be established in the same way it would in non-locked in people showing cognitive impairment.

Diachronic autonomy of individuals with ALS in CLIS

In contrast to other cases of CLIS, the inexorable nature of ALS enables anticipating CLIS. People with ALS could therefore prepare decisions in advance. However, advance consent to PAS is not sufficient as they need to be able to carry it out themselves. One could ask whether, from an ethical standpoint, the presence of an advance directive on PAS in these ALS patients in CLIS would permit to accept some uncertainty in capacity evaluations via BCI.

This means that if people with ALS have a very stable wish to die if they ever enter CLIS, a lower standard of communication could be deemed acceptable because they just reiterate a previously stable wish. Indeed, ALS might be the only case where the reliability of communication and the training of algorithms on BCI data is feasible because individuals can still communicate well before CLIS. This might give more certainty and credibility to the evaluation of capacity and communication of choice via BCI later.

Physical control as willed bodily movement via BCI and self-administration

Even if capacity could be established in some, if rare, cases of ALS, physical control of the intake of the lethal substance must also be established as this remains a major safeguard for autonomy in self-administered PAS. Prima facie there seems to be a plausible case to treat BCI control and physical control the same because legally, responsibility for misconduct via BCI is attributed to the person using the BCI (Bublitz, Wolkenstein, Jox & Friedrich, 2018; Thompson, 2019). Bringing about harm or damage to other persons or objects with the use of BCI does not exempt the person who used it from legal ramifications. Physical control, tort liability, and criminal responsibility are innately linked and that, if users of BCIs (e.g. patients in CLIS) are liable for actions committed against others, they should be able to be responsible for acts against themselves (e.g. PAS), too. Rainey and colleagues (2020) have argued that while foreseeability and issues of control distinguish BCI actions to some extent from conventional actions, in cases where "there is a clear reason for the use of a BCI technology for assistance or owing to disability, it appears easy to assimilate this kind of action". For the context of control via BCI in CLIS, the reasons for its use are clear: there is no other option for deciding about one's end of life. On this view, end-of-life decisions enacted via BCI are the only option of patients in CLIS, if one does want to uphold patient autonomy.

BCI and PAS – some caveats

We have argued that in some specific cases of CLIS, namely in patients developing CLIS in the context of ALS, PAS via BCI could be an option in the future, depending on developments in BCI technology. As the technology is still in its infancy, we have not specified what kind of BCI should best be used for PAS in CLIS.

This sensitive topic demands two caveats. The first is that enabling the option of PAS in CLIS, does not mean that life is not worth living in CLIS. Indeed, there are reports of a good quality of life for people in CLIS who were able to communicate via BCI (Holz et al., 2015). Even though this study was limited to five participants, it was an important proof of principle (Johansson et al., 2017). The option for PAS should therefore rather be understood as part of several strategies to increase autonomy and enable choice concerning a good life in CLIS. With this option for PAS, among others, it might indeed be reasonable that more people with ALS enter CLIS – and do not feel forced to die before their quality of life declines to a point where they do not wish to live any longer. Indeed, they would not be forced to decide for PAS or against invasive ventilation before they develop (completely) locked-in state, especially if quality of life may still be maintained there. It has been postulated that quality of life of people in CLIS might be even improved due to the availability of end-of-life options like PAS, as the choice provides some control over the current situation even if they do not wish to die (Colborn, 2020).

The second caveat relates to the state of BCI research. While there have been reports of established independent use of BCIs in ALS (Wolpaw et al., 2018), the research field has also had a major research ethics scandal with the research misconduct committed by Niels Birbaumer and colleagues on exactly the topic of communication with paralysed ALS patients (Chaudary et al., 2017; Chaudary et al., 2019; both publications have been requested to be retracted by the German Research Foundation). Not only does this leave open the question of effectiveness of establishing BCI communication in these patients, it will also most likely have undermined trustworthiness for intricate decisions via BCI at the end-of-life for patients and their families. Robust and replicable research procedures would therefore need to establish the viability of BCI communication before it could be applied in the context of PAS or WT.

Conclusion

In the future, assisted dying policies based on self-administration will be faced with the question whether competent people in CLIS should be enabled to end their life via BCI. We have argued that in some cases of ALS this might be ethically viable. Enabling patients to end their life even once they have entered CLIS might, paradoxically, prolong and uphold their quality of life.

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Chapter 8.

Blurring Boundaries at the End of Life in Home Care

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Blurring Boundaries at the End of Life in Home Care

A Look at Germany, Switzerland and the United Kingdom in the Light of Care Ethics

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Abstract

This chapter discusses blurred boundaries in home care at the end of life. Firstly, an overview of support structures for informal caregivers, i.e. family and friends, in Germany, Switzerland and the United Kingdom is provided. From the perspective of Care Ethics, three blurred boundaries are identified: 1. between being an informal caregiver and a non-caregiving close person, 2. between formal/professional and informal caregivers, and 3. between caring for others and self-care. The first blurred boundary relating to the role of the informal caregiver is especially crucial in dementia. With a slow progression, the line between being primarily a relative and being an informal caregiver becomes blurred. Formal and informal care in the Swiss case is blurred due to the employment of informal caregivers. The last blurred boundary lies within the informal caregivers themselves: being continuous caregivers, they are sometimes in need of care themselves. Based on that, a discussion of possible modes of support is provided: awareness promotion, supportive structures and an attitude of Care Ethics, which includes trust and responsiveness in relationships. The change from dyadic care relationships to care networks is encouraged.

Keywords:

informal care, self-care, dementia, care ethics, blurring boundaries

1 Introduction

Demographic change in Western societies has been exerting considerable influence on the health system as a result of the increase in the overall ageing population. Due to demographic change, the dependency *ratio* of older people in need of help to younger people providing this help is changing drastically. It was already foreseeable in 2005 that "the current and future provision and maintenance of a broad-based and differentiated range of social and health-related services as well as the supply and demand for nursing care in Germany and other countries will be influenced by critical demographic and labour market trends" (Meyer, 2005, p. 10, translation by the authors). Informal care by family and friends represents an essential pillar in the care landscape. Economically and socially, informal care is "a basic condition for the functioning of the organised health care system" (ter Meulen & van der Made, 2000, p. 257). In Germany, more than three quarters (76 % or 2.59 million) of all those in need of long-term care (LTC) are cared for at home, of which 1.76 million are cared for exclusively by their relatives (Destatis, 2018). In the United Kingdom (UK), the monetary value of the informal care provided equals between £57 and £100 billion (62–109 billion Euros) in professionals' working hours per year (Houses of Parliament 2018).

An estimated 60 % of all care in European countries is provided informally (Genet et al., 2012, 63). Informal caregivers are defined by the Organisation for Economic Co-operation and Development (OECD) as "people providing any help to older family members, friends and people in their social network, living inside or outside of their household, who require help with everyday tasks" (OECD, 2017, p. 208). This help at the end of life (EOL) usually covers all activities of daily living (ADL), including instrumental (e.g. household chores, shopping) and basic (e.g. self-care tasks, nursing) ADL. Informal caregivers, especially in EOL situations, also, for example, administer medication, maintain a tracheostomy or dress wounds. The latter tasks in particular explicitly demonstrate the blurring of boundaries between professional/formal and family/informal care (McDonald et al., 2017). Questions of responsibility emphasise the Care Ethical approach of self- and other-care. The conflicts between self and other care occur against the background of care work and are understood as boundaries that get blurred within informal caregiving.

Consequently, the aim of this international comparison is to examine informal caregiving and its blurring of boundaries against the background of Care Ethics. Our analysis highlights three particular blurred boundaries of informal care:

1. between having the role of the informal caregiver and not having it,
2. between informal (family) and formal (professional) care, and
3. between caring for others (other-care) and needing care for oneself (self-care).

This warrants explication. After providing a descriptive comparison of support structures in Switzerland, Germany and the UK, chapter 3.1 sets out how blurred the process of becoming an informal caregiver in the context of dementia is. In chapter 3.2, the boundary between informal and formal care is discussed. More specifically, the employment of Swiss informal caregivers by the state blurs the status of informal care. Lastly, informal caregiving is not as one-sided as the term caregiving suggests. The person giving care, this chapter maintains, is also most likely in need of receiving care. This chapter, therefore, weaves a tight narrative of how the role of informal caregivers is blurred in relation to certain other roles: the role of a non-caregiving other, the professional role and the care recipient's role. The cases in chapter 3 serve as reference points and are discussed in the context of Care Ethics. Care Ethics is instructive to highlight and cohere blurred boundaries, especially in the tension of self- and other-care as well as collaboration and co-operation between professionals and informal caregivers. The article particularly emphasises trust as the underlying ethical attitude of caregiving. It points to the necessity of the formation of care networks and highlights the need to increase public and political awareness of informal caregiving.

Our analysis is integrated into the international and interdisciplinary discussion on nursing ethics and professional ethics.[1] The specific situation of double-duty caregivers (Ward-Griffin et al., 2015) and volunteers engaging with terminally ill patients and their relatives (Sévigny et al., 2010) is beyond the scope of this article.

2. Legal and theoretical background

A comparison of the support structures of Switzerland, Germany and the UK is instructive, as these countries each follow a welfare state regime (Zigante, 2018, p. 14). They face a great demographic and societal challenge: people are living longer and the

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[1] Both aspects are particularly discussed by Schnell and Dunger, Seidlein and Salloch in this edited volume.

population of 65 years and over is growing faster than any other age group (United Nations 2019). As public resources for LTC are scarce, a 'care gap' is created and informal caregivers subsequently step in (Zigante, 2018, p. 23). The three countries have concurrently set up different support structures and pursue distinct policy strategies.

2.1 Comparison of support structures in Germany, Switzerland, and the United Kingdom

In Germany, the situation of care *recipients* has improved since the introduction of the two LTC Strengthening Acts (PSG I & II). They specifically guarantee insurance benefits and extended support in ADL. With the aim of reconciling work and informal caregiving, the Home Care Leave Act grants employees paid absence from work to care for a close relative. The exemption can be full- or part-time (§§ 1, 3 PflegeZG). The act explicitly mentions the companionship of a close relative in palliative medical treatment as a legitimate reason to be exempted (§ 3 VI PflegeZG). The length of time employees can take off work varies depending on the number of informal caregivers. Each close relative of a care recipient in palliative treatment, for instance, can request a leave of absence from work for a maximum period of three months (§ 4 III 2 PflegeZG). These regulations are complemented by free courses for home care, pension contributions paid by nursing care insurance and further cash and non-cash benefits (Plöthner et al., 2019).

In the UK, policies vary between England, Scotland, Wales, and Northern Ireland. Similar to German law, they all aim to support informal carers. In England, this support ranges from practical to financial and includes the provision of a carer's assessment (Care Act 2014 (England), s. 10), security benefits and cash benefits directly to the informal caregiver (more detailed: Zigante, 2018, 25–27). By contrast, carer's assessments are not offered in Germany. Furthermore, in contrast to Germany, all acts and their associated strategies currently in place in the UK put more emphasis on the local authorities' duty to support informal caregivers (e.g. Carers (Scotland) Act 2016, p. 24). Moreover, the statutory right to take leave from work is the same for all employees, regardless of caregiving duties. In the UK, it is up to the employer to offer informal caregivers support and/or flexible working hours beyond the legal provision (Work and Pensions Committee 2018, 15–20).

The issue of informal caregiver support has gained more attention in Swiss social policy throughout the last ten years. From the postulate Seydoux-Christe (09.4199) demanding paid vacation for parents of severely ill children, more and more policies have focused on integrating informal caregiving with work (Bundesrat, 2014). Different federal support options currently exist. Short paid leave from work (up to three days) to care for sick children or relatives is supported, as this is seen as a family duty. Reduced work inevitably leads to reduced pay for leave longer than three days. Apart from the legal obligations of employers, support exists through disability insurance and so-called old-age and survivor's insurance. People in need of help with ADL can get a ‘helplessness allowance’, with a differentiation of three stages of ‘helplessness’. The amount of the allowance is higher in disability insurance than in old-age and survivor's insurance. People who are ‘helpless’ can also apply for an assistance allowance which is used to hire help (Bundesrat, 2014). This is, thus, part of a ‘cash for care’ scheme, where support is rendered indirectly (via the care recipient) to a (formal) caregiver. Finally, there are several – often cantonal (i.e. federal) – additional services and support structures which aim to improve informal caregiver coping. The later discussed case of the employment of informal caregivers (chapter 3.2) is one of direct support by the cantonal home care services, which can be seen as part of ‘cash for care’ policies but extends beyond financial payment.

2.2 Care Ethics and Care Work

Comparing the public support structures is instructive to reveal blurred boundaries on a legal and political level. On a normative level, informal care blurs the care relationship between physician and patient on which traditional biomedical ethics and professional ethics centre. Neither biomedical nor professional ethics take into account overarching societal or political considerations. They do not appreciate the role of a patient’s relatives or the informal carers. Therefore, it is through Care Ethics that blurred boundaries that come with informal caregiving can be mapped and held together.

Informal caregiving at the EOL is part of care work, understood as both family and professional activities linked to household, parenting, caring for someone and nursing (Leget & Kohlen, 2020). The demand of good care work for elderly and dying patients brings the issue of resources and responsibilities onto the political and scientific agenda. Questions of power, inequality and conflict meld the approaches of Care Ethics together with those of care work (Kohlen, 2018, 271).

Most approaches to Care Ethics "start from the notion that humans are intrinsically dependent on and interconnected with one another" (Stensöta, 2010, p. 297). Care is seen as the process of responding to a human's needs and, consequently, Care Ethics is needs-based and "seeks to maintain relationships by contextualizing and promoting the well-being of care-givers and care recipients in a network of social relations" (Sander-Staudt, 2019). Kohlen (2016) points out that despite the different aims of the debates between Care Ethics and care work, there are commonalities. Informal caregiving can be embedded in the concept of care work. This is understood as reproductive work, which Schwarzenbach defines as "all those rational activities (thinking about particular others and their needs, caring for them, cooking their meals, etc.) which go toward reproducing a particular set of relationships between persons over time" (Schwarzenbach, 1996, p. 102). However, this is usually unpaid work, especially if provided in the home environment. While reproductive work might be a more suitable approach to caring at the beginning of life, responding to needs observed at the EOL is equally essential.

3 The situation of informal caregivers in Germany, Switzerland and the United Kingdom – two cases

3.1 Informal caregiving in dementia: the blurred boundaries between becoming a caregiver, becoming aware of it and realising the necessity of support

This subchapter sets out to uncover the blurred boundary between exclusively being spouse, child, neighbour, gradually perceiving oneself as an informal caregiver and assuming this role. At an individual and interpersonal level, it centres mainly on observations from informal caregivers themselves and their relationship with their relative with dementia. Dementia is a neurodegenerative disorder affecting cognition. Symptoms of the disease can progress insidiously; as a result, relatives of people who develop dementia – if present – can transition to a caregiving role gradually, increasingly taking over daily tasks which can often result in full-time caring and nursing (Kurz et al. 2017).

The social image of dementia continues to be a negative one: people often associate dementia with an impairment of higher cognitive functions and the gradual loss of self-determination and autonomy which can evoke feelings of fear (McParland & Innes, 2017).

The life expectancy of people with dementia can vary between 5 and 20 years and depends on the age of onset, the type of dementia and the progression through each stage of dementia. People with Alzheimer's disease, the most common type of dementia, for example, live an average of eight to ten years post diagnosis, but, due to the nature of the disease, they are increasingly unapproachable for the accompanying carers. Nevertheless, people with dementia can still be reached on the emotional level for a very long time, even though the rational level is executed and can be addressed less and less (DGPPN & DGN, 2016). They constantly lose the capacity to relate reflexively to themselves and others. Nevertheless, characteristics of their personality which were once central for them as individuals become visible at one point or another. This is what Kruse (2017, p. 337) calls "*Inseln des Selbst*" ("isles of the self"). One can reach these 'isles' on the emotional level and by getting involved in the realities of the life of the person with dementia (Kruse, 2016).

In Germany, around 1.63 million people over 65 were living with dementia in 2016 (Statista, 2019). There are no current studies or figures in Germany on LTC for dementia. It is estimated that about two-thirds of people suffering from dementia are cared for at home by their relatives (Deutsche Alzheimer Gesellschaft e. V., 2016). In the UK, around 850,000 people were estimated to be living with dementia (Prince et al., 2014).

8.3.1.1 Comparison of the German and British National Dementia Strategy in the context of informal caregiving

The UK launched a major project in 2009 to raise public awareness of dementia and made the improvement of dementia services a national task: "Living well with dementia – A National Dementia Strategy":

> The aim of the Strategy is to ensure that significant improvements are made to dementia services across three key areas: improved awareness, earlier diagnosis and intervention, and a higher quality of care. [...] This Strategy should be a

> catalyst for a change in the way that people with dementia are viewed and cared for in England. (DHSC, 2009, 9).

The initiators identified 17 key points that are important for implementation, predominantly at a local level with the aim of improving the quality of care for people with dementia and promote a better understanding of its causes and consequences. The objectives included: “improving public and professional awareness and understanding of dementia”, “good-quality early diagnosis and intervention for all” and “good-quality information for those with diagnosed dementia and their carers” (DHSC, 2009).

This strategy has resulted in increased public and professional awareness of dementia, dispelling myths about the disease and easing feelings of fear and helplessness through education, information transfer or regional project initiation. Support services also benefit people living with dementia as well as their caregivers, encouraging them to take action and seek help early in the disease process. As a care network is established, mutual respect and dignity develops and the people with dementia are encouraged to articulate their needs as they face new challenges in everyday life (Heller & Schuchter, 2018; Kruse 2017). Regarding Care Ethics, the purpose of caring is to fulfil human needs and ensure the protection of dignity and the personal development of the individual (Kohlen, 2018).

In Germany, until the beginning of 2019, there was no national, standardised care for people living with dementia, unlike the UK’s national dementia strategy (DZA, 2019). The strategy currently in progress is being developed from numerous national projects[2]: mainly regional lighthouse projects, well-functioning networks and initiatives (e.g., projects of the Alzheimer societies, regional associations or health institutions) or nationwide alliances that are involved in a wide variety of dementia-related activities. Since 2012, the Federal Ministry for Family Affairs, Senior Citizens, Women and Youth (*Bundesministerium für Familie, Senioren, Frauen und Jugend*) and the Federal Ministry of Health (*Bundesministerium für Gesundheit*) have initiated a wide range of co-operation projects which aim at leading to the development of a national dementia strategy (DZA, 2019).

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[2] E.g. https://www.wegweiser-demenz.de/allianz-fuer-menschen-mit-demenz/die-allianz/nationale-demenzstrategie.html (accessed 18 July 2019).

Therefore, the topic of dementia should no longer only be dealt with at the level of the federal states: The responsibility should fall on the society as a whole in order to improve the individuals affected and their wider social network. More than a decade later than the UK, Germany is now establishing a strategy to combine the forces of many actors in a co-operative, binding and long-term process (DZA 2019). The differences between Germany and the UK become particularly apparent in the formation of public opinion. Knowledge of the demographic change and its effects have been obvious for more than ten years, but it has been only in the UK that this awareness led to public action.

3.1.2 What does informal care work mean in the context of dementia?

Public awareness and attitudes towards dementia in Germany were explored in the German Employees' Health Insurance Nursing Report 2017 (Klie, 2017). The authors of the study discussed the conditions for a good life with dementia. The social and emotional integration of a person with dementia was identified as an important factor. There is a desire "to preserve the dignity and self-determination of the person as far as possible" (Klie, 2017, p. 23; translation by the authors). Regarding social interaction, the interviewees wanted more respect for people with dementia and more appreciation for the caregiver. Dissatisfaction is expressed about a lack of financial support but also the desire for more help from professional nursing services has been raised (Klie, 2017). These desires for rewarded (emotional, social and financial) care and the recognition of the usually invisibly time invested have been regarded as central pillars of Care Ethics for many years (Kohlen, 2018).

The boundaries between becoming aware of being an informal carer, assuming the role as a caring relative and the self-image as an informal caregiver are blurred, particularly in the context of dementia. This blurred boundary regularly comes with a decrease from self-care to other-care or even leads to another blurring boundary: the one between caring for another person and needing care for oneself. It is particularly in this context that the consequences and measures a person takes for individual mental hygiene are of great importance (more European studies can be found in chapter 5). In most cases, the carers – especially in the home-based care of a person with dementia – are often late in revealing their own situation and needs (Klie, 2017). It can be difficult as an external observer to fully appreciate and provide appropriate care arrangements (Bruker & Klie, 2017). In this context, Care Ethics aims to highlight the inevitability of self-care.

> It includes caring for one's own health, education and individual needs. Care work for oneself is necessary at the job, in the family and with friends as well as in other areas in order to perpetuate one's working capacity. (Kohlen, 2018, p. 268; translation by the authors)

Caring relatives are part of the wider environment of people affected by dementia and can make a significant – supportive and stabilising – contribution to their loved one's ADL by adapting to their diminishing abilities (Engel, 2007). Friends and neighbours are rarely involved in the case of dementia, because caregivers often feel ashamed of the behaviour of their dementia-affected relatives. Therefore, both run the risk of being subject to social isolation (Meyer, 2005). This circumstance contradicts the principle of Care Ethics, also regarded as relational ethics, which refers to the fact that people are existentially dependent on each other (Kohlen, 2018). Thus, people with dementia – due to their symptoms – and their caring relatives increasingly withdraw from social life. Caregivers have less time for their family, their social environment and their job due to the increasing amount of time spent caring.

> In addition to time pressure and years without leisure, rest and the pursuit of personal interests, the conditions under which care is provided for at home cause reduced social and family relationships, the loss of friendships and the feeling of loneliness. (Müller et al., 2008, p. 10; translation by the authors)

The changes that dementia brings about for the family may threaten the familiar interplay of individual family members and upset the balance of their relationships (Wilz, Adler, & Gunzelmann, 2001). In Germany,

> the family [...] continues to be *the* central institution which provides instrumental and emotional support for older people [...] and home care is often still considered

> to be a private responsibility to be shouldered by the closest family members. (Meyer, 2005, p. 11; translation by the authors)

Dementia makes the patient increasingly dependent on third parties. Due to the emerging care situation, a change of roles takes place within this family system, and new responsibilities have to be identified. The spouse "with whom [...] [before the illness] a mature relationship was maintained" is in constant need of care (Kruse, 2017, p. 363; translation by the authors). Regarding one's own children, there is a "reversal of the old 'power relations' [or] to [...] [their] 'parentification'" (Wilz, Adler, & Gunzelmann, 2001, p. 27; translation by the authors). Everything changes – the child suddenly makes decisions for the parent or parent-in-law concerned, gives instructions, praises, punishes, washes and takes over many more tasks. This new circumstance can lead to the parent developing feelings of shame and guilt, aggressive behaviour but also to a special devotion to the child (Wilz, Adler, & Gunzelmann, 2001, p. 27).

The carer's lack of autonomy to schedule their own day often leads to the impression that one cannot satisfy everyone's needs, leading again to feelings of guilt and stress (Meyer, 2005). In addition, constant supervision by the care-giving relative is necessary due to the increased urge for mobility of many dementia patients, which conveys a feeling of being tied down (Müller et al., 2008; Seidl & Labenbacher, 2007). The Data Report 2018 of the Federal Statistical Office summarises the expenditure on care for people affected by dementia as follows: "The presence of dementia leads to an increase in the degree of care with otherwise identical conditions" (Destatis, 2018, p. 331; translation by the authors). The average daily care of a person with dementia takes 4.5 hours (Gräβel & Addabo, 2012). Pinquart and Sörensen (2002) find that the care for people with dementia is mentally more stressful than for those who do not suffer from dementia. The most common mental illness is depression, which, in turn, is considered to be the strongest predictor of dementia (Ismail et al., 2017; Schäufele et al., 2007; Wilz et al., 1999).

The assumption of nursing care and support pushes most elderly people to the limits of their ability to cope with the frequent effect that the carers themselves fall ill and become "hidden patients" (Meier et al., 1999, p. 86). This, in turn, could eventually result in the primary informal caregiver becoming a co-client (Twigg, 1989, p. 59–60). Being a co-client means becoming a client of the health services as a result of giving care, as this can affect the care-giver's physical and/or mental health in a burdensome way. At worst, they also become patients, dependent on formal care – changing sides and facing boundaries between formal and informal care again – because the level of self-care has not been sufficient to cope with the strain. In an attempt to prevent becoming a co-client in these highly emotional surroundings, informal carers often need to maintain a balance between their own psychosocial or economic needs and the needs of the care-receiver. Their self-care and the embedment in a whole care network is of special importance (see chapters 4.1 & 4.2). Nevertheless, boundaries both between formal and informal care, self- and other-care are still highly blurred to date in Germany.

In spite of all the burdens that nursing care can entail mentioned above, it must be mentioned at this point that informal caregivers also often experience "mental enrichment and fulfilment in the process of nursing" (Kruse, 2017, p. 357; translation by the authors). These positive aspects must not be forgotten or overseen in conjunction with all the negative feelings.

The findings listed underline the special relationship and value of caring relatives and friends. It is always important to ensure the integrity of both parties – caregiver and care recipient – to avoid improper advantage when offering support. The 17 key points of the British strategy make clear that all parties involved in the care relationship, the person with dementia, the caring relative(s) and the care network surrounding both, must be considered and treated equally in terms of information, knowledge transfer and support. The perspectives of all those involved should be carefully explored and given equal consideration – reciprocity is the keyword here (Kruse, 2017).

It has become increasingly evident that informal caregivers need emotional, financial, practical and social support, especially against the background of blurred boundaries between self-care and other-care. A specific option of support is discussed in the next section.

3.2 Blurring the boundaries of family caregiving: the case of employment of family informal caregivers in Switzerland

When family members become informal caregivers, they are often in need of some form of support. Different international support options, for example, 'cash for care' policies, have been laid out above. Internationally, one measure of support for informal caregivers in Switzerland is employment through home care services (*spitalexterne Pflege; Spitex*), which is unique for Switzerland because it is restricted to 'cash for care', but includes pension and insurance benefits, as well as supervision and working in a multidisciplinary team. Conditions for employment of informal caregivers are: a) a basic formal nursing qualification, b) working age, and c) providing only LTC. However, it only subsumes basic (*Grundpflege*) not advanced nursing care (*Behandlungspflege*), as only the former can be reimbursed by health insurance. Contracts are usually time-limited and end when the care recipient is admitted to a nursing home or dies (Von Ballmoos & Despland, 2010).

This model of support is unique internationally, because it is provided directly to the caregiver. Most models of support, for example, payment through local authorities in the UK, only offer indirect financial support, i.e. the care recipient receives an allowance which is used to hire care (Zigante, 2018; see chapter 2.1). As outlined above, such a model exists in Switzerland as well. Employing informal caregivers, on the other hand, is a form of direct support: employment is between the informal caregiver and the cantonal home care services. This is similar to the Structured Family Caregiving programmes partly existing in the United States but also notably different: these programmes provide literally "cash for care", while employment arguably extends beyond receiving payment (even though getting paid might be the most important component). Denmark has a very similar model to Switzerland: in the former, informal caregivers can be employed by the municipalities. In contrast to the Swiss model, informal caregivers in Denmark are paid for more than 10 hours a week and continue to receive pay even if the person is in a hospital or nursing home (Zigante, 2018). Furthermore, the Danish caregivers are neither included in a multidisciplinary team nor receive training. Although international comparisons exist, the Swiss situation is unique. Data from 2008 indicate that 40 spousal or family caregivers were employed in six Swiss cantons (Von Ballmoos & Despland, 2010). Under 0.5 percent of home care services staff is made up of informal caregivers (Von Ballmoos & Despland, 2010). Thus, overall employment of informal caregivers is marginal. Nonetheless, the topic has been covered by media outlets and local initiatives due to the moral issues allegedly raised by the employment of informal caregivers[3]. From the perspective of Care Ethics, it will be argued that while the employment of informal caregivers in Switzerland blurs the boundaries of family and professional care, family care remains undervalued.

3.2.1 Is employment permissible from a Care Ethics perspective?

Care Ethics is essential to understanding these moral issues because it provides a particularistic approach suitable to analysing different moral situations, which, nonetheless, is centred on human relationality. In this subchapter, we use Care Ethics as a normative standard to lay out moral issues in the employment of informal caregivers.

Most moral issues of the employment of informal caregivers pertain to a certain understanding of caring: i.e. the view that family care belongs to the "world of the gift [that] moves through a continual affirmation of bonds, based on responsibility, trust, and gratitude and premised on our capacity for wholehearted attachment" (Hochschild, 2013, p. 168). From this point of view, if employment in some way infringes on the nature of the care or the attitude of the caregivers within familial relationships or if it is commodified to the extent of exploitation of the informal caregiver (Blaser, 1998), employment becomes an unacceptable form of support. However, employment becomes impermissible only if it is an empirical fact that care is meaningfully changed by the employment of informal caregivers. Indeed, Stacey and Ayers (2012) describe that payment is perceived by family caregivers as a norm violation. The norm violated is that family care should be a gift and, hence, unpaid.

Understood in this way, one could assume that one essential feature of Care Ethics might be in opposition to the acceptability of employment of informal caregivers: Care Ethics demands that caring attitudes are upheld (Kittay, 2001). Employment of informal caregivers, on the other hand, might 'crowd out' the caring attitude to care, as care is rendered for instrumental reasons: to receive payment and pension benefits as a co-worker of "agencies [which] work in parallel with the informal sector" (Twigg 1989, 58).

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[3] For the political controversies in the Swiss town of Uster, see https://www.sp-uster.ch/vorstoesse-details/189.html (accessed 27 September 2019).

This goes hand in hand with the worry that care might not be provided in the care recipient's best interest: clearly, providing unwanted care falls short of having a caring attitude. These worries do not seem to be empirically substantiated. Empirical research generally suggests that welfare state support complements family support or even strengthens it (Daatland & Herlofson, 2003; Künemund & Rein, 1999; Motel-Klingebiel & Tesch-Römer, 2006). By contrast, crowding-in just effects are also plausible. If informal caregivers have the option of employment, they might more often take up informal care for a relative or friend as they can choose between two types of paid employment. Welfare state support, such as employment of caregivers, could then work as "encouragement of family help through formal service provision" (Motel-Klingebiel, Tesch-Römer, & Kondratowitz, 2005, 866).

Clearly, this does not say anything about the effect that payment has on caring attitudes. However, it is at least plausible that caring attitudes are sometimes hindered by the demands of care, namely the financial costs (Feinberg et al., 2011) caused partly by the reduction of working hours. Consequently, some form of institutional support is needed for caring attitudes to come into effect.

3.2.2 Can employment be care?

So far, we have argued that employment is not generally impermissible from a perspective of Care Ethics. This is because a) the assumption that employment affects care and caring attitudes in a negative way is unsubstantiated and b) because the consequences generally for informal care (to increase the provision of informal care, i.e. to crowd in, or to decrease the provision of informal care, i.e. to crowd out) can be positive or negative. At this point, we do not have any reason to believe that care and caring attitudes are in any danger due to employment. Indeed, we may ask whether employment can actually have positive consequences for informal caregivers. Employment could possibly constitute care for informal caregivers. This would treat informal caregivers not only as care-providers and co-workers but also as potential care-recipients, i.e. co-clients (Twigg, 1989, p. 59–60), which is to say that family caregivers are in need of care themselves. These needs are not the same as the immediate care work they themselves provide. It is more an understanding of needs than the capacity to benefit (Asadi-Lari, Packham, & Gray, 2003; Wright, Williams, & Wilkinson 1998). In the healthcare context, this means that "if health needs are to be identified then an effective intervention should be available to meet these needs and improve health" (Wright, Williams, & Wilkinson, 1998, p. 1311). Employment, from this point of view, could be an effective intervention regarding self-care. Family and informal caregivers can be in a precarious situation, not only financially (Feinberg et al., 2011). Additionally, family caregiving can be lonely (Vasileiou et al., 2017) and overly demanding if caregivers are not trained for the tasks they are required to perform (see chapter 3.1). Employment could be a potentially useful tool here to ameliorate the situation of informal caregivers. Benjamin Miller (2016), for example, has argued that legal institutions could and should be designed to care. This is part of a wider push for a *Public Ethics of Care* (PEC; Stensöta, 2010). According to Helen Stensöta, a PEC makes two promises. Firstly, that through PEC, "the government makes *a statement that relations between people are seen as intrinsically important*" (Stensöta, 2010, p. 299, original emphasis), and secondly that PEC "provides a framework for how care-related services and provisions tasks can be performed in the best way", which is "possible through the value of responsiveness" (Stensöta, 2010, p. 299). Employment, from this perspective, could be a caring legal administrative device if it values caring relationships and is responsive to the caregivers' needs. It can also help to hold the blurring of the boundaries between caring for others and caring for oneself at bay.

The Swiss model of employment only partially meets the standards set in these promises. Employment clearly does not make a statement of the importance of caring relationships. That is because employment is only offered to informal caregivers who have at least basic nursing training and are of pre-retirement age. Hence, this excludes older informal caregivers without professional training. This perpetuates a professional service view of care that is not compatible with the wider reproductive notion of care. Care Ethics demands that all care work is seen as work, not only professional care limited to certain tasks. The first promise is not fulfilled by the Swiss model of employment.

The latter only partly fulfils the second criterion of responsiveness. It provides informal caregivers with some additional income and with pension years, which they would not receive without employment. Additionally, in most cantons, informal caregivers are closely supervised and work in a team, even though they are only responsible for one care recipient. This helps with loneliness and the financial costs of caregiving. Nonetheless, the Swiss model falls short of providing care in some regards: financial needs are only minimally met, as employment is compensated maximally for ten hours of care per week. This is because only basic care is compensated by the home care providers. The latter, while cantonal, are largely paid by the Swiss health insurances, which limits the care work to a narrow understanding of health-related care work. Consequently, care is only relevant if it relates to the fulfilment of tasks related to health care. As outlined, this is an understanding of care clearly incompatible with Care Ethics.

4 Managing blurred boundaries: Awareness promotion, supportive structures, and Care Ethical attitude

The two cases and the legal and theoretical background particularly reveal three different blurred boundaries.

(1) On the level of the individual informal caregiver, lines are blurred between becoming an informal caregiver, becoming aware of being an informal caregiver and being recognised as such. Moreover, and transcending the scope of the present article, the recognition as an informal caregiver might lead to blurring boundaries between private and public space, which also is reflected in the British National Dementia Strategy.

(2) Boundaries between formal and informal caregivers are especially blurred if they share tasks and responsibilities, such as feeding or mobilization, therefore, complementing or supplementing each other (Jacobs et al., 2014, p. 58). This is especially the case where informal caregiving at home coexists with professional home care. While the Swiss case shows that paying informal caregivers for their care leads to a blurring of boundaries between family and professional caregiving (Genet et al., 2012, p. 10), the blurring of boundaries between work and home life, especially if informal caregivers are dependent on the good will of their employers as it is in the UK, needs to be researched further (Martin-Matthews & Phillips, 2008).

(3) Finally, both cases repeatedly unfold that informal care can – and often does – lead to a blurring of boundaries between care for another person (the care-receiver) and care for oneself. This aspect is particularly important in the light of Care Ethics and care work (chapter 4.1).

While care-givers' self-care is insufficiently addressed in all three countries (Germany, UK and Switzerland), the international comparison implies two opposing trends. On the one hand, the case of Switzerland shows that boundaries are not blurred enough. While employment could be designed as a caring legal administrative device, the current Swiss model of employment is not acceptable from the perspective of a PEC because it upholds the boundary between informal and formal care. This is to say that while informal caregivers' work is now valued within the labour market for the first time, only part of it is. Reducing informal caregivers' work to professionals' tasks falls short of making a decisive difference for informal caregivers. Rather, it upholds the strict division between family and professional care. While it might be a well-meaning start to value informal caregivers' work, it does not follow through and potentially excludes the major part of care work performed by caregivers above retirement age or without professional nursing training. Furthermore, the Swiss model continues to see 'care work' as a mere fulfilling of medical and nursing tasks, excluding the vast segment of 'reproductive work' outlined above in section 2.2. Consequently, it lacks the remuneration of equally important caring tasks, such as doing grocery shopping, preparing meals, interacting with the care recipient and looking after the latter's mental and emotional well-being. For a thorough responsiveness and recognition of informal caregivers' contributions to the functioning of the 'formal' health-care system, 'care' has to be more comprehensive. On the other hand, the case of caring for relatives with dementia emphasises the importance of countervailing blurred boundaries between the informal caregiver and the care recipient. Due to the increasing isolation of elderly couples with one partner being affected by dementia, providing care for others often comes with insufficient care work for oneself. Such boundless care might only be revealed if informal caregivers themselves become co-clients or even patients.

Although pointing to opposing trends, both cases illustrate the crucial significance of embedding informal care into a larger context and care network: the extended family, formal care services, but also health and social institutions, legislation and politics. At the same time, future policies have to emphasise and ensure informal caregivers' self-care.

4.1 Between care work for others and for oneself

The State of Caring report from the UK (Carers UK, 2018) represents the necessity of institutional support for caregivers. Most of the participants mention that they are neglecting themselves, are worried about their personal safety and feel socially isolated. The process of social isolation and becoming aware of oneself as a caring relative usually takes place slowly. Consequently, the boundaries between 'giving a hand', 'caring' and 'nursing' also blur slowly. In addition, relationships get out of balance due to caring: Within marriage, one-sided dependency leads to a loss of balance and of interaction at eye level; in the parent-child relationship, roles are reversed. Children suddenly decide for their parents and take over tasks that were previously clearly the responsibility of the parents (Lawson, 1987, p. 275).

Thus, practicing self-care is especially difficult for informal caregivers as they are involved in a strong social network and can hardly leave their newly acquired role (more aspects are discussed in chapter 5 including perspectives also from the European South). It is not surprising that they often put forward the wish of respite, a short period of relief from care in the form of outpatient day care or a joint stay of an informal caregiver and care receiver in a health resort (Plöthner et al., 2019). This wish has to be answered with sufficient offers that enable caring relatives to achieve comprehensive mental hygiene in order to find a balance between their own needs and those of the person to be cared for (Colombo et al., 2011,p. 97–103). It is up to national legislation to adhere to the European Charter for family carers that addresses in its eighth principle the "right to respite": "This is a fundamental necessity to be met in terms of support, occasional and/or emergency assistance, substitution services and/or top-quality temporary care centres during various periods of time depending on needs (holiday, rest, health, etc.)" (COFACE Disability, 2017). In the UK, day and respite care are already recognised by the health-care policy as being important "to keep things going for family and patient as long as possible" and are seen as an appositive service to in-patient care (Lawson, 1987, p. 277). Day care hospices, for example, are an important institution of respite structures for informal caregivers. While day care hospices have been an integral element of palliative care services in the UK since the 1990s (Higginson, 1993), Germany is just starting to establish day care hospices[4].

In addition to political and institutional developments related to supportive structures for informal caregivers, researchers such as van Kempen et al. (2012) point out that informal caregivers expect continuing psychosocial support and a patient-professional relationship. Furthermore, need for positive and respectful conversations and care decisions concerning the care-recipient's well-being is identified (Schwenk, 2017; Stolee, Zaza, & Sharratt, 2014). This would enable a continuous building of trust (see chapter 4.3) and the formulation of task profiles and reflection spaces which would contribute to successful co-operation (Schwenk, 2017, p. 241ff).

The case of Switzerland deals with at least some of these blurred boundaries, as the employed family caregivers frequently meet their interdisciplinary team and can share their experiences and burdens. Nevertheless, it is crucial to embed the care dyad into a whole network of care to address all aspects and hold up some boundaries between the informal caregiver and care recipient. Taking Care Ethics seriously, care work does not only tackle the people affected by care (giving or receiving), but also responsibility has to be taken over by politics and society at large.

4.2 Collaboration and co-operation, or taking advantage of blurred boundaries

Professional caregivers can be seen as one of the most important resources for informal caregivers in the home care setting. Involving professionals could be a relieving factor for all people involved in the care interaction, particularly at the EOL. When strong emotions, such as disgust or fear, are present in the interaction with the care recipient, professional caregivers can take care for others. One might think of situations where the boundaries of the body become blurred. Mutual support becomes significant especially at the EOL when aggressive tumours can destroy the body (Kaiser et al., 2019; Lawton, 1998). Informal caregivers often become overwhelmed in emergencies and even during routine care. On the one hand, they do not have the professional knowledge necessary

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[4] The foundation of the first German day care hospice was laid on 1 November 2018: https://www.presseportal.de/pm/103154/4100872 (accessed 27 September 2019).

for advanced nursing care (Hetue Hill, 2019, p. 123), and, on the other hand, they are involved in a much deeper emotional relationship with the care recipient.

Furthermore, the high need of training and education highlights the importance of co-operation and collaboration between professionals and informal caregivers. In cases of emergency or in the context of death and dying, for example, a lack of information can lead to fear and distress for both caregiver and care receiver. This can be resolved with stronger co-operative networks between formal and informal caregivers (Lund et al., 2014). This need for shared information arises within various studies (Crotty et al., 2015; LaVela et al., 2016; Lund et al., 2014) and is seen as a mutual aim to which both professionals and families have to contribute.

The findings about the co-operation and communication between professional and informal caregivers – a significant further blurring of boundaries between formal and informal care – demonstrate the need for a wider understanding of supportive structures for informal caregivers, as discussed in the case of employment of family informal caregivers in Switzerland (chapter 3.2).

4.3 Trust: The underlying Care Ethical attitude

The self-image of an informal caregiver depends strongly on the possibilities of support offered. They often reveal their own situation and needs too late. Lack of early support results in their transition into a co-client. In order for Care Ethics to be implemented more widely, reciprocity and responsiveness between the informal caregiver and professionals has to come to the attention of future policymakers to initiate practical interventions. This reciprocity is inevitably based on trust. Boundaries between formal and informal care can only be blurred successfully and without causing additional distress if all people involved trust one another.

Kittay highlights trust in the context of Care Ethics. She associates her experiences as a mother of a disabled daughter to her work as a philosopher: “[…] given […] the physical or mental incapacity, the trust invested in the [professional caregiver] not to abuse [the] power over the charge [by the physical or mental incapacity] is enormous” (Kittay, 2001, p. 561). She points out the power inequity within the care relationship and the necessity of a trusting relationship. Focusing on the relational aspect of care can be helpful to establish the responsibility and supportive role of institutions and professionals and emphasise the importance of both professionals and informal caregivers in the home care setting.

The example of trust shows some of the difficulties that arise when informal caregivers are made to suddenly share or hand over their care work to a professional. Consequently, being aware of the dynamics and boundaries of professional care towards family care is essential to support informal caregivers adequately. Again, a Care Ethical approach can offer normative guidance to circuit boundaries and highlight the comprehensiveness of care. As the family is "a social institution with a particular history and structure" (Tronto, 2010, p. 161), care for a family member is also of particular importance. Hence, institutions and professionals have to comprehend the individuality and diversity of care to meet the needs and preferences both of the care recipient and their family. On the one hand, this goal can be reached if professionals understand the caring process in the home care setting as a co-operative process where informal caregivers and care recipients are included with their personal and individual experiences and knowledge. On the other hand, this has implications for both institutions and homecare:

> […C]are institutions have to think about the nature of the caring process as a whole in order to guide their actions. This requirement does not only demand that the 'needs' of the 'customers' come first but also that the needs of care workers, the allocation of responsibility and proper assessment also happen within the organization. (Tronto, 2010, p. 162)

Consequently, good and just care can be provided only if caregivers, whether they are informal caregivers who form part of the 'institution family' or professional caregivers who form part of a caring institution, involved in decision-making processes are encouraged and involved due to personal responsibility and are perceived as persons with needs and wishes. Getting to the heart of the issue, Kittay (2001, p. 560–561) writes that "[…] to advocate for my daughter without also advocating for those who are entrusted with her wellbeing is at once unjust and uncaring toward the caregiver".

5 Blurred boundaries of care: a European perspective

The three compared countries all support caregivers through a welfare-state regime. However, as we have shown by analysing the blurred boundaries of informal caregiving in the UK, Germany and Switzerland, caregiving is a cultural and welfare context-dependent activity. There is a variety of factors influencing the caregiving work done every day. The legal system enables substituted decision-making in the case of dementia, or the support services of the welfare state, provide, for example, state-funded pay for caregiving.

Generally, welfare state regimes in OECD countries have been classified according to the typology by Esping-Andersen (1990) as liberal, conservative, or social democratic. In this typology, the UK is a liberal, while Germany and Switzerland are conservative types of welfare state. Confirming this model, the UK would provide more individual means-tested support while Germany and Switzerland would support families more in providing care.

There have been several criticisms of this typology, most importantly that it is gender-blind and neglects the European South (Michoń, 2008). Michoń (2008) shows how the familisation-defamilisation dichotomy has been introduced as a gender-sensible alternative to the aforementioned typology. Following this typology in our analysis, we have studied two welfare systems with a certain degree of familisation (Germany, Switzerland) and one more defamilisied welfare state (the UK).

The degree of familisation of welfare systems influences how the blurred boundaries identified in our analysis play out in different European countries. Generally, the European South has been identified as a paragon of a familistic welfare state. Italy, Greece, and Spain are conventionally seen as Mediterranean countries where care is typically provided by families. However, also Eastern European states show high degrees of familisation in both policy and cultural value (Szelewa & Polakowsky, 2008). Therefore, within the European context these states provide a worthwhile comparison to the states discussed so far.

A key factor is cohabitation. Barbosa and colleagues (2020) found in the SHARE study that "in the Central, Eastern and Southern European regions, co-residential caregivers aged 50+ are predominantly women, with the percentage of female co-residential caregivers in the Eastern and Southern regions being considerably higher than in the Northern and Central regions".

Assuming that in Mediterranean states with a high degree of familisation, caregivers and care-recipient as well as other family members cohabitate more regularly, selfcare might become harder to achieve due to more obligations of care to others. The boundary of self- and other-care is especially blurred under these circumstances.

In parallel, the boundary between becoming a caregiver and being a non-caregiving relative or significant other is more blurred when there is an expectation by the state that care is provided within families. Generally, becoming a caregiver in the context of dementia is similar in most European contexts. For the typical process of becoming a caregiver has been described in an Estonian narrative study: "The interviews revealed that relatives of people with dementia noticed a deterioration of cognitive functioning years before the care began. They did not seek help, however, because they did not suspect that the impairments could be the symptoms of dementia. Family members wrongly assumed that the peculiar behaviour of their loved one was a normal sign of ageing" (Varik et. Al, 2020, p. 450). Clearly, this is an universal feature of a progressive neurodegenerative disorder. Nonetheless, the ability and way caregivers provide care is influenced by the welfare context.

However, even in welfare states generally supporting families (with a high degree of familisation), support can come in fractured ways. Wulff and colleagues (2020) state that "Italian caregivers often lack support in terms of formal services, home-care and professional training from the government". Here not familisation but the general unavailability of support services influences informal caregivers to immediately take over the caregiving role. In their comparative paper, Wulff and colleagues point out: "Italian caregivers are more likely to live with their care recipients, consequently spending more hours in caregiving compared to Swedish caregivers. As they find less time for themselves and are more exposed to physical, emotional and financial constraints, they are more likely to experience psychological distress". It is therefore possible that the most basic distinction in the European context regards the general availability of support services, and that it is secondary if this welfare state shows a high degree of familisation. However, the perception of the availability of services by affected families might be more important than the availability of services themselves. Koukouli and colleagues (2020, p. 7) have shown in their study that

> [f]amilism and gender were two predisposing factors found to affect perception of services. These seem to be important in the Greek culture and they may be in any other collectivistic culture where the individuals are interdependent, value, and rely on the family. Therefore, perception of services may not be influenced by their adequacy and availability *per se*, but also by the cultural values of the perceiver, and should be taken into consideration in relative studies.

Here, familism is not understood as part of a way policies are structured around supporting the provision of care – if it is dependent on family structures or independent of it. Rather, familism represents how care is structured culturally around the family and consolidated by familistic public policies.

There are two caveats to the discussion of familism. First, familism has majorly been studied within the context of childcare provisions. Hence, when welfare states are described as familistic, this might not hold for the provision of family care for older adults. Second, the existence of familistic welfare-systems has often been equated with the existence of cultural family values and family structures. Especially the Mediterranean states in the South of Europe have often been seen as paragons of family-based care provision. However, this is more complex than the notions of familistic or collectivistic suggest. In their aptly named article “The Myth of the Mediterranean Family” Calzada and Brooks (2013) describe how the familistic focus on the Southern European countries only holds up in comparison to specific Northern countries. These limits and the partly too focused approach need to be taken into account when comparing the – apparently – same phenomenon across different countries.

6 Conclusion and Outlook

Formal and informal care, other- and self-care are not mutually exclusive but interdependent. It is this care triad of care receiver, formal caregiver and informal caregiver that is essential for a single care recipient and their well-being. Nevertheless, formal and informal caregivers and care recipients are part of a community and society as a whole. It is a characteristic of care that it is relational, blurs boundaries and often requires more than one counterpart. This nature of care and the difficulties addressed highlight the necessity to think and act in care networks, which are based on trust and mutual understanding.

However, as demonstrated by the two cases of Germany and Switzerland, the national support structures and legal regulations in both countries still regard informal caregiving as an isolated phenomenon which is mainly tackled on the microsocial level. Although the family is still regarded as the central institution which provides instrumental and emotional support, the changing family systems that have occurred over time bear a growing risk of comparatively early institutionalisation (Genet et al., 2012; Zigante, 2018). This phenomenon must be counteracted effectively with alternatives and attractive supportive options.

In the vein of calling for a more comprehensive social, political and legal responsibility in the care network, this analysis also emphasises the importance of professional ethics to include the general public and health-care professionals. This means that a care policy involving the public ensures that care work is not solely delegated to a (vulnerable) group of people who are left alone with the responsibility of caring but rather to an attitude that shapes policymakers, administrative services, public employees and civil society (Eichner, 2015, p. 94–98).

Caregiving must continue to be brought into public awareness in order to improve the social image of caring professions and caring in general. The people affected and their relatives can benefit through information and knowledge transfer and wide-ranging support. A thorough change requires a “System Thinking” (Werhane, 2003, p. 89) that integrates all health-care stakeholders. Among these are not only care recipients, formal caregivers and informal caregivers, but also the government, educators and researchers, the wider community, professional associations and many more (Werhane, 2003, p. 84). Society can be challenged through Care Ethics “to rethink the nature and purpose of politics and the political vocabulary of justice, freedom, privacy, and the like in terms of what is necessary for promoting and sustaining good personal care" (Engster & Hamington, 2015, p. 1).

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Chapter 9.

Hopelessness in palliative care for people with motor neurone disease

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Hopelessness in palliative care for people with motor neuron disease — conceptual considerations

Christopher Poppe

Abstract

Hope and its absence, hopelessness, are seen as essential in palliative care for people with motor neuron disease (MND). A primary measure in psychological research on hopelessness in people with MND is the Beck Hopelessness Scale (BHS). This scale can be understood as being conceptually based on the philosophical standard account of hope, which understands hope as an intentional expectancy. This essay argues that this is a misconstruction of hopelessness in palliative care. Rather, pre-intentional hope is essential for palliative care of people with MND. Pre-intentional hope enables the formation of intentional hopes and is intrinsically relational. Finally, it is argued that the absence of pre-intentional hope should not be subjected to psychiatric diagnosis, for example in the form of demoralization disorder.

Introduction

One might observe a child and wait until one day he manifests a hope; and then one could say "Today he hoped for the first time." But surely that sounds queer! Although it would be quite natural to say "Today he said 'I hope' for the first time."

Ludwig Wittgenstein, Zettel

In palliative care, hope is commonly seen as of utmost importance (Kylmä et al., 2009). Hopelessness of patients is to be avoided at all costs. Nevertheless, there are illnesses where on first sight there is very little to hope for. In the case of motor neuron disease (MND), there is no known cure and the disease progresses inexorably. Thus, it is a paradigmatic case for palliative care (Borasio et al., 2002). The World Health Organization defines palliative care "as an approach that improves the quality of life of patients and their families facing the problems associated with life-threatening illness" (World Health Organization, 2019). Hopelessness is a major restriction to this quality of life.

In this essay, I will argue that one influential conceptual account of hope misconstrues hopelessness for palliative care. If we understand hope as a wish or an expectancy towards the future, people with MND might be seen as hopeless. If a person with MND for example would have a hope for a cure, she might become hopeless as she realises that the degree of probability of her hope is zero. She would have lost a hope. While this understanding of hope might seem naive and it is clear that hope should be understood in greater terms, I will show that it has been influential in psychological research on hopelessness. The concept of hope as expectancy is then contrasted with hope as a "pre-intentional state" (Ratcliffe, 2013). In the conclusion, I compare absence of hope as a pre-intentional state to demoralization disorder.

What is the standard account of hope?

Hope is a notoriously vague concept. A common definition of what has been called the "standard account of hope" (Blöser & Stahl, 2019) states:

> "A hopes that p" is true iff "A wishes that p, and A thinks that p has some degree of probability, however small" is true. (Day, 1969)

Understood this way, there are various hopes of different probability a person with MND might have: the hope that a cure is found in one's lifetime, the hope that the disease might progress slowly, the hope that one dies a good death and is remembered, and many others. Some of these hopes might prove to be false hopes given their low probability. Similarly, there has been a distinction drawn between optimism and hope, where optimism is understood as an unrealistic expectation.

To see what the use of the concept hope in palliative care is, it is worthwhile to take a closer look at the instruments used to measure it or, to be more precise, the absence of it. For the sake of brevity, I will focus on one instrument commonly used to measure hopelessness, which is used in the context of MND (e.g. Plahuta et al., 2002): the 20-item Beck Hopelessness Scale (BHS). Originally, the BHS was deemed to measure pessimism, i.e. negative expectations about the future (Beck et al., 1974). The naming of the BHS has thus been misleading and any critique of it should encompass that the originally envisioned construct measured is only partially hopelessness, even though it has often been used and understood as such. The standard account of hope is most prevalent in the BHS. Items #12, #6, #8, and #13, among others (see Table 1), measure negative expectancies about the future and thus the absence of hope on the version of the standard account. Here the low, negative expectancy of a wished-for outcome is seen as the absence of hope, as hopelessness (even though we may only rightfully call it pessimism).

Table 1: Standard account of hope in the BHS [8]

BHS item no.	**Item statement [(Beck et al., 1974)]**	**Relation to the philosophical standard account of hope**	**Probable mismatch with palliative care, esp. amyotrophic lateral sclerosis**
#12	I don't expect to get what I really want.	**Intentional hope** "I hope that p, where p is to get what I really want".	Depends on the really wanted good, if we assume that death is not really wanted it is **probable mismatch**
#6	In the future, I expect to succeed in what concerns me most. (inverse item)	**Intentional hope** "I hope that p, where p is to succeed in what concerns me most."	Success is possible if it relates to main concern of palliative care to have a *good death*. **Probable mismatch**
#8	I expect to get more of the good things in life than the average person. (inverse item)	**Intentional hope** "I hope that p, where p is to get more of the good things in life than the average person".	Might be cynical in the context of having an incurable, terminal disease. **Probable mismatch**
#13	When I look ahead to the future, I expect I will be happier than I am now. (inverse item)	**Intentional hope** "I hope that p, where p is to be happier than one is now".	Depending on the assumption that death could provide relief; **probable mismatch**

Thus, if a person agrees with (or, in inverse items, disagrees with) these items, he or she would score higher on the BHS and be more hopeless. As some researchers suggest a cut-off of 9 'to be indicative of suicide intentions' (Kliem et al., 2018), even as little as the misconstruction of four items might make a difference if hopelessness is overestimated in the context of palliative care. Hence, there is strong reason to believe that measuring intentional hopes, i.e. expectations, might not suit objectives in palliative care. To strengthen this argument further, we can ask whether loss of intentional hopes leads to hopelessness.

Loss of intentional hopes

If a person with MND would be to lose all these intentional 'hopes that p', would this person then be hopeless? In the literal use of this word, certainly, but it is not sure whether this literal use is qualified for the use in palliative care. We just do not know whether the features that are often psychologically associated with hopelessness, i.e. depression, despair, and demoralization, are indeed present in the absence of intentional hopes.

Similarly, Matthew Ratcliffe has argued that the absence of hopes, understood as valued expectancies, is a too simple understanding of hope. On his account, "it is possible to lose all hopes (…) and yet retain another kind of hope" (Ratcliffe, 2013). His phenomenological version of this kind of hope is a "pre-intentional state", where "certain kinds of intentional state, including intentional hope, are intelligible" (Ratcliffe, 2013). The existential feeling of hope therefore precludes all other version of hope, i.e. it allows oneself to form positive expectancies. It is more procedural than outcome-oriented. With reference to Jonathan Lear (Lear, 2006), Ratcliffe continues to describe this sort of hope as *radical hope*, which he understands as "a kind of general orientation or sense of how things are with the world, in the context of which intentional states of the kind 'I hope that p' are possible" (Ratcliffe, 2013). It is a state, where expectations can happen to begin with, rather than some form of expectancy as an intentional state. We can further compare the state of absence of intentional hopes to indifference. When nothing is expected, this would render a person hopeless. Nevertheless, it might just be that this person is indifferent to the future or denies the importance of thinking about it. Indeed, present-centredness might be of worth for a person in palliative care. Thus, to lose pre-intentional, radical hope is different from the loss of all intentional hopes.

For now, the origins of pre-intentional hope remain unclear. There might the case that people with strong religious beliefs and faith might be more likely to have pre-intentional hope in face of a terminal disease. This would go hand in hand with the empirical observation that the wish for hastened death is restricted by religious belief in patients with MND (Stutzki et al., 2012). Another factor, I hold, might just be as likely and compatible to this observation: it is the relational nature of pre-intentional hope. To have faith, on this view, would be to feel in relation to a higher deity. Nonetheless, even on a secular view, relationships are fundamentally important at the end of life. Social support and closeness to next-of-kin are crucial in enabling pre-intentional hope. In the language of object-oriented psychoanalysis, it is the attachment to others, to our primary caregivers, that shape the beginning of life (Tan, Zimmermann, & Rodin, 2005). It is very likely that the end of life is equally shaped. Pre-intentional hope, on this view, could be seen as basic trust in the world, shaped by our relationships with other persons in it.

The loss of pre-intentional hope

Loss of pre-intentional hope, on this version, would be more dramatic than any loss of intentional hopes, as measured by the BHS. It would be closer to a state of despair, depression, or existential angst. It is also closer to what has been called demoralization syndrome (Kissane, 2000) that consists of "existential distress […] characterized by feelings of hopelessness and helplessness due to a loss of purpose and meaning in life" (Robinson et al., 2016). Furthermore, the rationale "for its importance as a syndrome stems from the associated desire for hastened death that accompanies such feelings of hopelessness and meaninglessness, raising issues of competency in the context of rational or physician-assisted suicide" (Robinson et al., 2016). Here, we see how a faulty conception of hope in palliative care might be misleading; not only would, as I have argued, suicidal ideation be overestimated, but also the desire for a hastened death would be medicalised and subjected to the realm of psychiatry. If hope would be conceptualised as an expectancy, a person with MND might be unrealistically be seen as hopeless because she does not expect much from her future. It might still be that this person has some form of pre-intentional hope left, which would slip by the BHS, for example.

Rather, I hold, pre-intentional hope has to be attained by not only the person affected with MND, but also in relation to next-of-kin, nurses, and all other healthcare practitioners involved. This in line with the infamous dictum of palliative care: that people just want to die a hastened death, if there is a lack of (good enough) palliative care. We can assume that the authors of demoralization syndrome would argue in a similar vein. In this argument, it is of special importance not to prescribe hope: It is not the case that all people with MND should have pre-intentional hope, even if their relational circumstances might make pre-intentional hope attainable.

If one were to prescribe hope to people with MND, this would come close to pathologising suffering (Rich, 2014). Anyone who would be pre-intentionally hopeless, on this view, would be pathological and in need of a prescription of hope. Similarly, not all people have to want a prolonged death, even if there is good enough palliative care. There are cases where pre-intentional hope just is unattainable and these cases have to be accepted, rather than being subjected to a psychiatric diagnosis. In some cases of MND, there might simply be nothing to attain or sustain so-called radical hope. It seems to be rather absurd to demand that these dying patients have to undergo further psychological treatment for demoralization disorder. Rather, everything possible should be done to comfort them and to meet their needs in the last weeks to days. On this view, good palliative care encompasses a non-specific treatment for demoralization syndrome. However, it might not be effective for every possible case.

Conclusion

As I have shown in this essay, current research employing the BHS misses an important point about hope. Rather than conceptualizing hope as a form of expectancy, hope should be understood as a pre-intentional state. Pre-intentional hope is a vital concept in the palliative care for people with motor neuron disease. However, pre-intentional hope was not laid out substantively in this essay. We can nonetheless say that pre-intentional hope is of relational nature. Furthermore, I have tried to argue that loss of pre-intentional hope is different from demoralization syndrome. Finally, for the context of MND we might have to reformulate Kant's famous question of "For what may I hope?" to "How can I hope?".

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Chapter 10.

Fair access to assistive technology? Gaps in transition from invalidity to old-age insurance in Switzerland

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Fair access to assistive technology? Gaps in transition from invalidity to old-age insurance in Switzerland

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Main text

Amyotrophic lateral sclerosis (ALS) is a fatal neurodegenerative disease leading to increasing immobility, disability, and death within 3-5 years from disease onset (Kiernan et al., 2011). In two thirds of patients, age of onset is between 50 and 70 years (Caroscio et al., 1987). Considering demographic changes in Europe, this means that ALS will become significantly more frequent in ageing populations (Arthur et al., 2016). Due to the progressive nature of this illness, patients with ALS need access to different sorts of assistive technology and aids, ranging from wheelchairs to electronic communication devices and others from the field of neurotechnology (McFarland, 2020). Facilitating appropriate access to these technologies is an important element to ensure good quality of care for people who suffer from ALS.

In this Op-ed, we argue that in the care for people aging with ALS and diagnosed after retirement, there are gaps in equal access to assistive technology in Switzerland. These gaps appear when the age of onset of the illness or its progression coincides with the transition from eligibility for invalidity insurance (IV; Egli et al., 2021) to old age insurance (AHV). IV is a social insurance scheme covering loss of wages for people who have some degree of disability due to medical inability to work before retirement. It aims to enable insured people to conduct an autonomous and independent life and encourage companies to employ them. AHV is the social insurance scheme that provides retirees with a minimum income (which is supplemented by cash benefits from occupational pension and private savings/reserves) and basic support. Since these two different social insurance schemes guarantee different level of benefits, the mere fact that ALS is diagnosed just before or just after being eligible for one or the other insurance scheme has a profound impact on access to assistive technology (Rickli, 2016).

This represents a shortcoming with regard to justice, a key principle in research on the ethics of use and development of assistive technology (Wangmo et al., 2019). Justice in terms of equal access to healthcare is indeed a fundamental principle for people with ALS and their families, as well as with respect to disability and ageingaging in general (Rickli, 2016).

To illustrate our point, consider these two short fictional cases:

> 1. Ruedi got diagnosed with bulbar ALS at age 64 and received assistive technology (electric wheelchair, communication aids) through IV before retirement and transitioning to AHV as a social insurance scheme. He is now 67 and, due to rapid progression of ALS, his need for assistive technology increases even more. Although being already retired and now covered by AHV, Ruedi is guaranteed vested rights (Besitzstandsgarantie; EDI, 1978, Art. 4) to have the same level of access to assistive technology as he had before through IV (Landolt, 2014).
>
> 2. Maria got her diagnosis of spinal-onset ALS after a year-long diagnostic odyssey at age 65 when she was already retired and thus covered under AHV. She got access to assistive technology through AHV. She is now 67 and AHV only provides her access to a non-motored wheelchair every five years but not any other technology (EDI, 1978; Landolt, 2014).

It is clear from these examples that Maria is worse off than Ruedi with regard to access to assistive technology. At the same time, also in Ruedi's case his transition to AHV allows him to retain same level of benefits that he received when insured under IV, which may in the future not be adequate to address his worsening condition.

The difference in access to assistive technologies between IV and AHV generates two questionable gaps. Firstly, it is paradoxical that Maria and Ruedi are both 67 years old and they both are living with the same diagnosis, but their access to assistive technology – which is a precondition for them to participate in public and private life - is substantially different only due to the time of diagnosis (for Ruedi just before passing to AHV and for Maria just after). In Maria's case, if a private foundation's support or own financial means are not available, she might have to live without communication or advanced mobility aids. Due to early(ier) diagnosis, Ruedi is still covered by IV and has thus guaranteed access to a broader range of assistive technologies. Early diagnosis of ALS can thus paradoxically be seen as beneficial since it allows easier access to assistive technology.

Secondly, even when access to technologies is guaranteed through IV the problem of delayed access to these benefits remains for people with ALS. Indeed, there is a year of waiting time before invalidity pensions and financial aid (Hilflosenentschädigung, Assistenzbeitrag) can be received through IV. This waiting time is normally used by the administration to ensure that insured person is unable to work, but this can be illogical in the case of ALS patients where the disease progresses rapidly. For someone with ALS with a rapid progression, this waiting time means that – even in the best of situation where diagnosis comes during IV eligibility – financial support and payment for personal assistance might come too late. While this does not extend to assistive technology and aids, it provides further evidence of how these policies are unfit to meet the needs of people with ALS and their families.

This general clash between the rigidity of some aspects of the Swiss social security system has also been pointed out by Rickli (2016), who concluded that the „institutional transition in status from 'disabled' to 'old' at a bureaucratically relevant point, but an arbitrary stage in a person's disability history, leads to a confrontation with the social security system within a new, yet less entitling category of citizenship" (p. 16). We agree with this statement. In ALS the aforementioned paradoxes generated by the transition from IV to AHV lead to a problematic unequal access, and, evaluating this from the perspective of engaged bioethics (Scully, 2019), to a difficult situation for people with ALS and their caregivers.

While the gap in transition from IV to AHV might be unjust to people with disability in general, in ALS there exist feasible steps to ensure access to technologies enabling participation in public and private life, and therefore upholding quality of life. People with ALS should be treated by administrative institutions as hardship cases, toward whom there needs to be more attention (cf. Egli et al., 2021). As part of these hardship cases the waiting year normally necessary before receiving some benefits under IV should be skipped.

These issues of access to benefits are also internationally encountered for people with ALS and their families. Recently, the non-govermental organization Marie Curie and the Motor Neurone Disease (MND, a synonym for ALS) Association in the United Kingdom have called for a change of law regarding the current ‘six-month rule’, which allows terminally ill people to cut red-tape for accessing welfare benefits only when they have less than six months to live. The organization calls to modify this rule and allow every terminally ill person, e.g. people with ALS, to cut red-tape, even when the life expectancy is longer than six months. Marie Curie organization puts it bluntly: “Dying people shouldn’t have to waste time worrying about getting the support they need.” (2021). In ALS, every month counts, and waiting for a year in Switzerland for benefits is too long. some of the benefits obtained through IV is too long. Both welfare benefits through IV and access to assistive technology through AHV therefore lack adequate considerations of the condition and needs of people with ALS and their families.

Recently, the bioethicist Joseph Stramondo argued for a “right to assistive technology” justified by compensatory justice for the disadvantage created „by society’s dominant cooperative scheme and the violation of [disabled people’s] right to equality of opportunity that such disadvantage entails“ (Stramondo, 2020). Understanding the concept of justice as such is one ethical argument in favour of changing the gap related to the transition from IV to AHV in relation to assistive-technology access by ALS patient. Even if this argument from compensatory justice is seen as not enough, a further ethical justification in favour of remedying this gap can be provided by the most extreme forms of egalitarianism which only aims to level the playing ground but does not make excessive demands on justice. For example, luck egalitarianism (Bickenbach, 2016) would demand that access to assistive technology should be the same regardless of the fact whether the illness is diagnosed *just before* or *just after* the transition from IV to AHV. Indeed, the time of diagnoses, as exemplified in the two fictional cases, can be seen as a form of ‘bad luck’ upon which the patients have no control.

With increasing availability of innovative technology aids for people with ALS in the future – such as robot skeletons or brain-computer interfaces – existing gaps in access will only increase in size. Since these gaps can have a significant impact on the ability to participate in public and private life, it is problematic that broad access to assistive technology remains dependent on pre- or post-retirement diagnosis of ALS.

Equality for people with disability has been enshrined into Swiss legislation (Disability Discrimination Act). While Switzerland has taken big steps to ensure that people with disability can take part in the workforce, for older adults with disability or those who transition to old age with disability, there are gaps in equality of access that can also turn into a form of age discrimination for this particular group.

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Chapter 11.

Discussion

11. Discussion

This cumulative thesis incorporates seven articles (published in or submitted to peer-reviewed journals) and one book chapter. By its very nature, the research presented in this thesis is a mosaic. In the following, first the different findings relating to methods A, B, C and D will be discussed in the form of a critical overview of the *Lay of the Land* (Kon, 2009). Afterwards, a stepwise integration of the empirical and normative elements of this thesis will be provided using Critical Applied Ethics (CAE; Leget, Borry, & De Vries, 2009). After this, future research directions are delineated, and clinical and policy recommendations are tentatively proposed as a conclusion.

In short, the main findings of this thesis are:

A. The systematic review synthesized evidence of quantitative, qualitative, and mixed-methods research on caregivers' needs in certain stages of caregiving trajectory and highlighted a dearth of evidence in certain phases (before diagnosis and after bereavement). The review found different needs relevant after diagnosis, across the caregiving course, and at the end-of-life. In the initial stage, the process of getting the diagnosis and responding to it formed crucial needs of informal caregivers. The synthesized needs across the caregiving course incorporated information needs on care provision and support available, emotional support for dealing with a terminal disease, an increased need for professional support with care at home and respite care, and needs relating to healthcare services. Further needs at this stage were financial support for the costs of supporting a person with ALS, dietary needs especially with PEG, and role, social, and other needs of informal caregivers. During the terminal stage, relevant information needs, needs with regard to palliative and professional care at the end-of-life, and needs regarding assistive technology and advance planning were present. Among the key conclusions of the systematic review was that future ALS caregiving research and practice needed to pay tribute to this differentiation.

B. The qualitative study of supportive needs of informal caregivers of people with ALS in Switzerland resulted in themes of support. Key findings were unmet needs regarding administrative demand, in contact with healthcare providers, while providing care at home and during the terminal phase. The importance of social support was highlighted from the perspective of informal caregivers. Healthcare professionals outlined availability and gaps in institutional support for informal caregivers, their own work as caring for informal caregivers, and general challenges in healthcare for families of people with ALS.

The thematic analysis on bereavement experiences and support highlighted sometimes emotionally complicated reactions to bereavement, sources of support, experiences of informal caregivers who adjusted to life without the person with ALS. Healthcare professionals reported on support they offered but also identified gaps in the available support. Bereavement support needs to be proactive from healthcare professionals; however, it requires goodness of fit to address those in need and not those who are adequately supported by informal sources of support.

C. The qualitative study using a grounded-theory approach theorised informal caregiving as a balancing act of different caregiving activities. Informal caregivers try to hold a steady state during the caregiving process. This process of holding of the balance is the organising element to four secondary categories which could disrupt or facilitate a balance. Organizing support could both shift the balance in favour or against the caregiver depending on the availability of support and being present could restrict or enable informal caregivers' activities. The categories of managing everyday life and keeping up with ALS further impacted the steady state.

D. Ethical, legal, and social issues (ELSI). Four separate ELSI were identified and explicated from a theoretical perspective. These ELSI were inspired by informal caregivers' remarks in the interviews or discussion in the literature. Most relate to conditions of end-of-life care for people with ALS. Brain-computer-interfaces were identified as crucial assistive neurotechnology which brought with it ELSI, for example, in exercising patient autonomy in different end-of-life decisions. Concepts of hopelessness in ALS make a difference in how people with ALS are assessed at the end of life. Blurred boundaries of informal caregiving were delineated at the end of life, and unequal access to technology needed to exercise autonomy in ALS was criticised.

11.1 Supportive needs in informal caregiving for people with ALS

In general, the findings of this thesis emphasise the distinct nature and social interrelatedness of needs of informal caregivers in ALS. Informal caregiving is a dynamic process changing during the caregiving trajectory and spanning the whole lifeworld of caregivers and people with ALS. While informal caregiving in ALS can involve medically complex tasks, it is not only a medical or healthcare process where professional support is needed but a rudimentary social process often carried out within the close dyad of the informal caregiver and the cared-for person with ALS. For each of the chapters, a critical discussion in the context of the whole of this thesis will be provided.

Critical discussion of the systematic review

Our key finding from Method A was that needs of informal caregivers are found at every stage of the caregiving course and different at each of these stages with multiple caregiving tasks involved. Some needs, e.g., information needs seem to be more important in earlier than in later stages of caregiving. Generally, pacing and planning of support for and involvement of informal caregivers according to the different caregiving stages seems warranted. International guidelines on the multidisciplinary care for people with ALS and their families underline this importance, especially with regard to early diagnosis, planning of care, and support (Andersen et al., 2011). Our research adds to these guidelines that multidisciplinary care not only has to be provided early, but also adapted to needs at the current caregiving and disease state. For example, discussions surrounding advance care planning should not be had when informal caregivers have been brought out of balance but when they have achieved a steady state again (e.g., a while after the 'shock of diagnosis'). However, often the disease state (e.g., the state of respiratory insufficiency) trumps the demands of the caregiving state, and care must be planned even though informal caregivers might currently not be ready for it.

The systematic review identified important gaps in research on informal caregiving in ALS. One of these gaps was bereavement care for informal caregivers. Since bereavement care is an essential part of palliative care according to the WHO definition of 2002 (Sepúlveda et al., 2002), this is a decisive shortcoming of the international literature. The qualitative research of Method B which used interviews with bereaved caregivers addressed this gap in the literature. From this thematic analysis of the interview data, the key conclusion was that healthcare professionals should adapt support after bereavement to the needs of the individual caregivers and monitor potential needs for further support (Aoun et al., 2017).

The methodological choice of narrative synthesis was, as already pointed out in the chapter, due to the different research designs in the included articles. However, in the future, this might change, and a substantial basis of evidence in the form of interventional studies and randomized controlled trials could exist. In that case, the narrative synthesis provided in this thesis could be complemented by a meta-analysis of measures of needs and interventions addressing these needs. In retrospect and due to the material shortcomings of the research field, a systematic review including the grey literature (policy documents, documents by patient organizations, guidelines to support informal caregivers) would have provided nuance to the analysis (Mahood, Van Eerd, & Irvin, 2013) as it generally influences outcomes of systematic reviews (McAuley et al., 2000). It could be the case that the academic literature in the field of informal caregiving research might only partially represent the knowledge available on the topic. This is especially the case as most actors providing support for informal caregivers (e.g., ALS associations and centres) are not necessarily academic institutions.

While the systematic review did provide a comprehensive narrative synthesis, it did not entail higher levels of evidence traditionally associated with the methodology of systematic review and meta-analyses. Our narrative synthesis therefore had the aim to provide an interpretation and critique of the existing evidence through a deepened understanding of supportive needs (Greenhalgh, Thorne, & Malterud, 2018). In turn, the qualitative research builds on top of this deepened understanding. Furthermore, as one of the conclusions of the systematic review was that the scope of interventions had to be widened from psychological interventions to psychosocial interventions, the research focus of Method B was laid on social supportive needs. Hence, the systematic review also took the function of preparing the qualitative study as we used it to both direct research questions as well as semi-structured interview guides.

Critical discussion of the thematic analyses

The qualitative research is providing a *Lay of the Land* (Kon, 2009) of supportive needs of informal caregivers of people with ALS. These findings add to an international corpus of knowledge but are especially important within the national context of fractured support and availability of palliative care throughout Switzerland.

Building on Method A, the qualitative research largely described supportive needs as a form of social needs. This holds for bereavement support, respite, low-threshold, and peer-support, but also extends to end-of-life care. The possibility and provision of palliative care at home in Switzerland and especially the availability of respite care through hospices or other institutions is a crucial gap outlined from the qualitative research. When it comes to informal caregivers providing palliative care involving medically complex tasks (e.g., administering morphine, caring for tracheostomy), these tasks need support from palliative specialists at home. Internationally, this thesis adds knowledge on support needed from healthcare providers, support at home, and support during the terminal phase.

Informal caregiving as a primarily social process fits well into new endeavours to start caring and compassionate communities (Abel, 2018). Indeed, as outlined in Chapter 8, support for informal caregivers should be arranged in care networks. In contrast, our qualitative research shows that some informal caregivers harbour privacy concerns against such networks, and that caregiving frequently remains a largely individual process, often within a marriage. Furthermore, in our sample a major part of informal caregiving was provided by female caregivers (daughters and wives), pointing to a gendered and cultural dimension of care. These dimensions, between informal caregiving as a private process within a family or a public process within a community, are clearly blurred. Nonetheless, this dimension was not focused upon in this thesis and leaves a gap for more research.

The discrepancies in the reports of informal caregivers and healthcare professionals are worthwhile to be considered. The concerns raised by some clinicians in our study about the autonomy of informal caregiving were not found in informal caregivers' reports pointing to the different perceptions of caregiving within a caregiving dyad and from the outside. This raises methodological as well as ethical concerns which were outside of the thesis's scope and have yet to be explored.

Critical discussion of the grounded theory-approach

Our theoretical understanding of informal caregiving in ALS entails a core category, holding the balance of a steady state. The core category is relevant in ensuring care and normalcy as the disease progresses until the end-of-life. It is the primary social process of informal caregiving from the perspective of informal caregivers and therefore guides decisions surrounding caregiving. On this understanding, if caregivers succeed in holding the balance, this is key to understand why some informal caregivers are able to provide care at home until death. We already discussed the limits of this theoretical understanding due to its flexibility in the chapter itself. Due to this flexibility, the theoretical understanding also connects well to the other parts of this thesis. It allows to generate new hypotheses regarding informal caregiving. For example, it is currently unclear whether in complicated grief in bereavement of informal caregivers the balance shifts after bereavement or whether it has already been shifted against the caregivers before bereavement. Theorising informal caregiving as achieving a steady state allows to develop multiple pathways for research and care for individual caregivers. Moreover, this dynamic understanding is less deficiency-oriented than theoretical understandings of caregiver burden or stress theory (Bastawrous, 2013). Indeed, it also allows to see positive or salutogenic experiences of providing care (Lin, Fee, &, Wu 2012). While this theoretical understanding remains vague this is necessary to deal with the heterogeneity of informal caregiving and disease in ALS.

Critical discussion of the conceptual analyses

The current thesis examined four ethical, legal, and social issues (Chapters 7, 8, 9, and 10). However, all the ELSI explicated in this thesis were only identified indirectly from the qualitative research part of this thesis. The explicated ELSI were raised by individual caregivers during the interviews and then theoretically explicated.

The analysis of end-of-life decisions and brain-computer interfaces (Chapter 7) took its cue from remarks by informal caregivers noting that their loved ones had gone too early due to the demands of assisted suicide and the people with ALS' fear of losing physical control over their fate. The analysis served the purpose to point out a particular puzzle in bioethics resulting from legal requirements at the end-of-life for people with ALS. This analysis centred on the individual autonomy of a person with ALS, however, as with the analysis of hopelessness and fair access to technology, these are also interconnected with relational autonomy of informal caregivers.

Informal caregiving at the end of life and its blurred boundaries were part of a European comparative effort trying to set informal caregiving within different political contexts (Chapter 8). In the interview study, informal caregivers had remarked on the possibility to get employment for caregiving, a possibility only existing in some cantons and already abolished in others. This led to a conceptual analysis of conditions for employment from the perspective of Care Ethics. We argued that employment of informal caregivers is permissible but in actual conditions falls short of constituting care for the caregivers themselves.

Theoretical discussions of hopelessness in the literature sparked an interest to write about its conceptualisation in ALS. This led to an understanding of hopelessness (Chapter 9) as an existential feeling that essentially is relational. Hopelessness is often seen as a psychological impediment to decision-making capacity. In that case, hopelessness is understood similarly to demoralisation rendering the person with ALS incapable to adequately decide on their options. However, hopelessness in ALS should not be measured as intentional hope, the hope for certain state of affairs. Understanding hope as pre-intentional allows to see hopelessness as an achievement by the caregiving dyad or network and leads to a less individual understanding of hopelessness in end-of-life circumstances. This conceptualisation of hopelessness therefore goes beyond intentional hope as it is typically assessed for people with ALS.

Lastly, narratives of informal caregivers in the interviews about losing further access to assistive technology and insurance benefits after entering retirement were grounds to write a short commentary to make the issue known to the professional public (Chapter 10). The availability of communication aids but also support with invasive ventilation shape the set of options people with ALS and their caregivers have at the end-of-life. Hence, decision-making at the end-of-life is influenced by social and political factors. From the social factors, the relationship with the informal caregiver stands out. It is likely that only if informal caregivers are well supported and able to handle caregiving practically and emotionally, they are able to provide the care until the end-of-life. Politically, policies regarding end-of-life decision-making such as assisted suicide as well as policies regarding the material conditions of caregiving are relevant.

Furthermore, the results of this analysis indicate that the nature of ALS with its foreseeable and inexorable fatality make decision-making unique.

11.2 Integrating the normative and empirical

This thesis entails a patchwork of research from multiple methods. Using the methodology of CAE (Leget, Borry, & De Vries, 2009), these mosaic pieces will be tried to put together to integrate the empirical and normative findings of this thesis. Necessarily, as part of the larger puzzle that is research, any integration is limited. These and other limitations are discussed subsequently. CAE has been introduced in Chapter 2 and in the following the five step-approach is outlined. However, not all the steps are equally studied in this thesis with most research belonging to the first two steps of CAE. The status of this research as the base of the pyramid is illustrated in Figure 1.

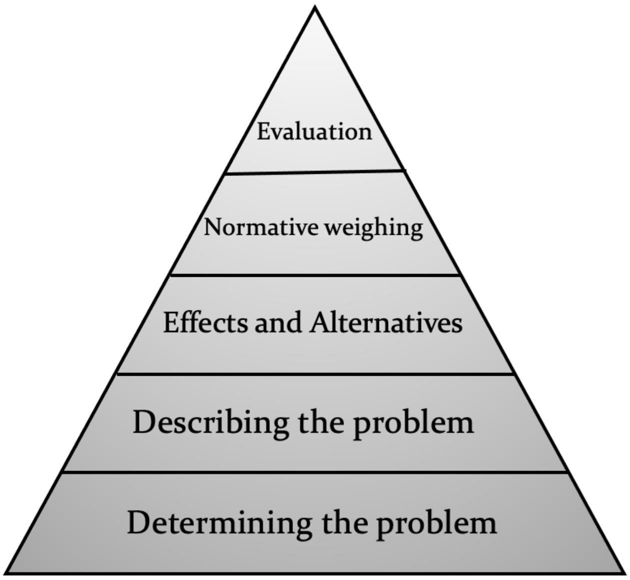

Figure 1: Relative evidence generated in this thesis

1. Determining the problem

Empirically, the supportive needs of informal caregivers are the most important findings and clearly demarcate problems for informal caregivers of people with ALS in Switzerland. These unmet needs are multidimensional and have been described in detail. Normatively, most of these unmet supportive needs can be described as problems of fairness and justice, broadly understood (see below in step 2).

Normatively, ethical, legal, and social issues were determined for the later stages of ALS. However, while several ELSI were determined for people with ALS (e.g., hopelessness, BCI-control), we did only partly identify ethical issues for informal caregivers from normative angles. One of these was the permissibility and conditions of employment of informal caregivers for their care from the perspective of Care Ethics. From the systematic review, one of these issues is the lack of knowledge available on bereavement support for informal caregivers which constitutes a shortcoming with regard to beneficent care for informal caregivers.

From the qualitative interview study, we could identify morally salient issues though, including autonomy in decision-making surrounding invasive ventilation in ALS, welfare state support, administrative demands for informal caregivers, relational autonomy in the relationship with neurocognitive impairments on part of the person with ALS, and potential impairment of decision-making capacity in ALS. Hence, both the normative as well as empirical analysis converge on the moral salience of decision-making at the end-of-life of people with ALS.

2. Describing the problem

The empirical research described several unmet needs in detail. Bereavement and generally supportive needs of informal caregivers have been described in detail, also leading to moral issues where there is a moral imperative for the responding institutions (e.g., administrative institutions burdening informal caregivers in bereavement) to act. Furthermore, a wide range of ethical issues have been directly identified by the qualitative research with informal caregivers and healthcare professionals (as outlined in step 1). While it would be worthwhile to have studied these ethical issues using further qualitative and quantitative research, this was not possible due to practical time constraints within this project. Rather, we have described moral issues from theoretical angles – the ELSI of Method D. In focusing on four different ELSI, we have described problems existing for individuals with ALS in exercising autonomous decision-making at the end of life (hopelessness, complete immobility) and relating to justice, broadly speaking (fair access, supporting caregivers politically in Europe).

There is strong evidence from empirical research as well as from the normative perspective that material conditions of caregiving (e.g., finances, aids) are a core problem for informal caregivers and hence, ethical considerations are often related to matters of distributive justice. These concerns relate both to the distribution of professional care (e.g., how much respite support is available from state-funded healthcare providers) as well as to the distribution of financial resources supporting informal caregivers (e.g., employment for their care or paid time off work). Distribution of financial resources is crucial because also low-threshold forms of social support for informal caregivers depend on them (e.g., setting up a café for peer meetings). Hence, lacking distributive justice, it is possible that social support through communities ameliorates lack of resources for informal caregivers and people with ALS.

From a normative perspective, the analysis of ELSI further shows that conditions of justice (e.g., distributive justice in form of access to technology) in turn influence conditions of care (e.g., the ability to communicate and thereby meet the needs of the person with ALS). Likely, this also influences the decision-making based on personal autonomy of the person with ALS (e.g., staying in touch with the caregiver upholds quality of life and diminishes wishes to hasten death).

3. Effects and Alternatives

To some extent, the supportive, unmet needs identified and described in this thesis are also the effect of existing policies. Certainly, this is true for some themes represented in the empirical research, for example, administrative demand after bereavement and during caregiving. Administrative demand is the effect of an administrative policy.

In our empirical study on bereavement support and experiences, we have discussed bereavement support between specialist intervention (e.g., palliative care physicians and clinical psychologists) and public health measure (e.g., Caring Communities). Here, there is clearly a lack of policies dealing with bereavement. Grief and bereavement are most likely seen as individual processes which do not demand public attention. However, this could be otherwise. For palliative care and neurology, this would mean taking a family-centred focus – that, even if the patient with ALS has died, the family is still at the centre of medical care, if needed. For public health and public policy, this would mean addressing bereavement as a communal issue. As we have shown the salience of social support for supportive needs of informal caregivers in our qualitative research, this would be in line with the demand for care within a network as outlined in Chapter 8.

Normatively, effects of policies are exemplified by Chapters 8 and 10 on the policy of employing informal caregivers and on the gap between invalidity and old-age insurance in Switzerland. Both these analyses are dealing with the effect of a policy or a possible alternative. Under the conditions outlined in Chapter 8, employment of informal caregivers could be a suitable policy to address financial needs of informal caregivers and provide recognition for their work. Chapter 10 argued for a special status of people with ALS within insurance systems and to grant insurance support as hardship cases. Here, the singularity of ALS as an often rapidly progressing terminal disease demanded special attention within welfare-state support. However, as these are the results of conceptual analysis, a comprehensive analysis of effects and alternatives of these policies is still lacking. It therefore primarily takes the function of pointing to problems and ruptured healthcare and welfare provision for informal caregivers resulting in ethical problems.

4. Normative weighing

As we have substantial evidence for unmet needs of informal caregivers, there is an open question what normative weight should come to the described problems and the possible alternatives. By itself, there is weight of our findings from a ‘Normative Force of the Factual’, a phrase borrowed from legal philosophy (Schauer, Bezemek, & Ladavac, 2019). In this thesis, this means that the availability of evidence on supportive needs of informal caregivers provides stakeholders with information on crucial problems for informal caregivers and respond to these accordingly. Furthermore, the earlier steps of CAE have outlined demands of distributive justice which have considerable normative weight in this thesis. Examples of this normative weight can be found in Chapter 10 where insurances as relevant institutions were addressed and urged to respond to unmet needs of access to technology as reported by caregivers. Furthermore, administrative demands and policies, as well as bereavement support as a public policy have considerable weight. The ethical issues from the qualitative research regarding relational autonomy and decision-making in the caregiving dyad and network are of utmost importance. Here, the normative weight first is important for further research in empirical bioethics (see 11.4 Future research).

While important for autonomous decision-making in ALS, the ethical and legal issue of brain-computer interface control over end-of-life decisions has considerably less weight than the access to brain-computer interfaces and other devices for communication at the end-of-life (Chapter 10). As outlined in the Chapter 7, this is because upholding communication is of greater importance than the possibility for an individual to act via brain-computer interface. Furthermore, with the exception of decisions for assisted suicide, upholding communication allows to communicate wishes and preferences of the person with ALS at the end-of-life.

Normative weighing naturally depends on the available evidence which in this thesis has only partly been provided for the ELSI. The most substantive normative weight therefore comes to the empirically described unmet needs of informal caregivers, more so than to the described ELSI as these were only ascertained from a normative perspective and not part of a larger empirical project.

5. Evaluating the Effects of a Decision

This thesis did not study interventions or policy decisions in any evaluative manner. However, it is possible to sketch out possible effects which could follow hypothetically from the research in this thesis.

Meeting the outlined demands of distributive justice in steps 1 and 2 would allow informal caregivers to better support the people with ALS at the end-of-life. It is very likely that with more resources for outside help, respite, and specialist palliative care at home, informal caregivers would be able to provide care at home until the very end. Moreover, from the empirically studied supportive needs of informal caregivers, several potential improvements of multidisciplinary care could be made. Standards of care should be harmonised throughout Switzerland. For example, advance care planning does not seem to be carried out similarly in all regions Switzerland highlighted by the different situations experienced by informal caregivers at the end of life. Generally, regional differences seem to persist. Specialist palliative care services and mobile palliative care should similarly be available to all informal caregivers throughout Switzerland. These services are vitally needed as at the moment informal care at home for people with ALS is without alternative as there is a lack of suitable infrastructure for the needs of people with ALS outside of the home. Therefore, an important effect is the visibility of the work informal caregivers of people with ALS provide.

Another potential effect is that the multidisciplinary care routinely provided to people with ALS pays more attention to bereavement of informal caregivers. This could be routinely implemented phone calls and checking up at certain time points after bereavement. In this way, healthcare professionals could screen for complicated grief of informal caregivers and activate potential sources of support.

While neuropsychological assessment of people with ALS is more and more routine practice, the effects of this on the caregiving relationship and network have not been highlighted. Therefore, an effect of this research could be that more attention is paid to the caregiving relationship and potential changes due to cognitive and behavioural impairments of the person with ALS.

As already pointed out, one effect of the research could be that insurances in Switzerland and other social institutions view cases of people with ALS and their families as hardship cases. This could have the effect of speeding up the time for administrative processes and lightening the administrative load of informal caregivers.

In the future, the development of brain-computer-interfaces could lead to widespread availability for people with ALS if these devices are of low cost or financed through insurances. This in turn could have the effect that the requirements for decisions made through a brain-computer interface demand more ethical and legal attention. These effects need to be studied in detail. Therefore, this step of CAE remains open, pointing to the need to discuss limits of the integration and the limitations in general.

11.3 Limitations

The integration of the empirical and normative research with CAE is limited. First of all, it did not address all steps equally due to the scope of this PhD thesis. Addressing all the steps of CAE would have required a larger empirical bioethical research project. Furthermore, the research questions were not primarily ethical questions which would have made an integration within CAE easier. Possible research questions for a more normative inquiry would have been, among others, "What does the state owe individuals who provide care to a dying person?" (cf. Reiheld, 2015), "What filial duties does a daughter of a person with ALS have?" (cf. Brakman, 1994). Hence, we have only determined empirical problems from the qualitative research without also identifying ethical issues from a normative angle. Additionally, these ELSI were therefore only supported by informal inquiry lacking a wider, substantial basis in qualitative research. Necessarily, this analysis is therefore at risk of having focused on normative issues as esoteric puzzles in bioethics – the esoteric puzzles which I found as salient. A ground-up qualitative research study on ELSI might have brought forward other, less esoteric, normative issues.

Beyond the challenges of CAE, the empirical and normative research presented in this thesis has limitations. Some of these limitations are intrinsically linked to qualitative research approaches as such and have been mentioned within the chapters. These include the small numbers of participant and the non-representativeness of the sample in general. Clearly, generalisations are not possible from qualitative research, and it is not the purpose nor the research question of the present thesis. Moreover, our qualitative analysis on needs of informal caregivers was largely descriptive due to the pragmatic focus of our research design. However, this pragmatic focus can also be seen as an advantage of this thesis providing hands-on evidence for social interventions (Smith, Bekker, & Cheater, 2011). Nonetheless, one chapter in this thesis provides a theoretical outlook on informal caregiving in ALS using a grounded theory approach. Furthermore, while this research primarily deals with needs of family caregivers, it is not situated our research with a sociology of the family (Gaugler & Kane, 2015) or intergenerational processes with changes of family norms and demographic change in aging societies, especially in Switzerland (Perrig-Chiello, 2012).

Additionally, my position within the research field might have focused the research on systemic health services research and less towards individually predisposing factors constituting needs of informal caregivers. One example is the common reports of guilt among informal caregivers (Martz & Morse, 2017). While we did ask for spiritual, religious, emotional, and psychological supportive needs, our semi-structured interview guides did only partly focus on the emotional and existential experience. While this is justified by the pragmatic approach in this thesis, the emotional and existential depths of informal caregiving might not have been adequately researched.

As already pointed out, the analysis of ethical, legal, and social issues did not arise systematically from the qualitative research but was sparked by remarks informal caregivers made in interviews. While this bases the analysis of ELSI on empirical ground, it is also likely that this gave too much room for focus on esoteric problems as problematised by Leget and colleagues (2009). Indeed, considering the state of modest availability of brain-computer interface technology for people with ALS, arguing for applicability in end-of-life circumstances may be closer to a thought experiment than to real-world decision-making. Yet, this might only presently be the case, and, if the outlined practical issues surrounding accessibility of this and other technology are ameliorated, it might indeed be necessary to consider such cases morally and legally.

11.4 Future research

Future research should aim at filling the gaps present from the overview provided by CAE and the general limitations of this thesis. For the CAE, a larger empirical bioethical project including all five steps and focusing on a particular policy issue would be a productive endeavour. For the limitations of our qualitative research, this non-representative research should be backed up with survey data from a representative sample of informal caregivers of people with ALS. Additionally, informal caregiving in ALS is already a very limited scope with ALS being a rare disease. Future research can take two strategies to address this limited scope:

1. **Specific topics** (e.g., non-related friends providing caregiving in ALS) could be studied by extending the scope of disease to similar neurodegenerative diseases such as Parkinson’s disease, Huntington’s disease, or other motor neuron diseases, e.g., Progressive muscular atrophy (PMA). While ALS is singular in its rapid fatality, caregiving in ALS shares features with these other neurodegenerative diseases. Future research could therefore study narrow topics by recruiting informal caregivers of different neurodegenerative diseases.

2. **Broad topics** (e.g., administrative burden in informal caregiving) should be studied in quantitative research designs with reliable and valid questionnaires to enable comparison. As mentioned, this should be facilitated with a representative sample. These broad topics could be addressed from a perspective combining empirical bioethics and disability studies (Garland-Thompson, 2017), including people similarly handicapped as people with ALS during the disease progression. This is even more so, as crucial bioethical topics, such as quality of life assessment are commonly shared and impact decision-making in clinical settings (Goering, 2008).

Our research identified potential ELSI which could be studied in a genuine empirical bioethical research program. For informal caregiving, one emerging ethical issue is neurocognitive impairment and its influence on relational autonomy in the caregiving dyad. If apathy and cognitive inflexibility are inflicting on the social cognition of a person with ALS, we know from the research already presented that this would impact the caregiver. Relational autonomy and the self-determination of informal caregivers are crucial bioethical topics which need further research.

Generally, decision-making in end-stage ALS is a worthy topic of research on its own, especially for invasive ventilation. At the end of life, invasive ventilation is a crucial decision from the perspective of patients, families, and the involved physicians (Borasio et al., 1998). However, in our qualitative interview study, Swiss physicians, and healthcare professionals reported unease in cases when patients requested invasive ventilation to prolong their life. Main factors stated are the burden of care for informal caregivers and the lack of support structures in Switzerland for ventilated patients. Furthermore, there is evidence that invasive ventilation is discouraged in ALS in Switzerland and France, as it is generally in Europe (Heritier-Barras et al., 2013). In contrast, in other welfare states, invasive ventilation in ALS is used more frequently. For example, this is the case in Japan. Almost a third of patients with ALS choose invasive ventilation to prolong their lives (Hirano & Yamazaki, 2010). Future research in ALS therefore demands a cross-cultural perspective, for example studying conditions of informal caregiving in different international contexts. Hence, future research also needs to address that care is provided within cultural and political contexts (Federici, 1975; Tronto, 1993, 2010).

For research, this leads to the conclusion that bioethical research on informal caregiving should be interdisciplinary. More precisely, the need to involve political and social sciences seems wanted. While within medicine there is nowadays an understanding of social determinants of health, there should be a similar research line concerned with the social determinants of informal caregiving. With the help of relevant stakeholders in public policy, this could lead to interventional research. This interventional research would be different from medical interventions often rendered out within a randomised controlled trial to achieve the gold standard of evidence (Meldrum, 2000). It would be a political intervention studied to see if this affects informal caregiving positively, for example, fast-tracking welfare applications of informal caregivers for assistance. This need not be large population-based studies in the beginning. Indeed, the fractured support within Swiss cantonal, municipal, and state organization of welfare could be a benefit as a cantonal pilot study could address needs-based administrative support of informal caregivers.

11.5 Conclusion: Clinical and policy recommendations

As a conclusion for these critical discussion points and future research lines, tentatively the following recommendations for clinical practice and policy action can be given:

- **Individualise needs assessment and re-assess during the caregiving course**. Different caregiving courses need to be taken into account clinically with individually coordinated support which needs to be available for all informal caregivers.
- **Standardise provision of palliative care**. Informal caregivers of people with ALS need to have equal access to palliative and supportive care throughout Switzerland.
- **De-medicalize informal caregiving research.** Considering that informal caregiver's needs were relevant in a social and political context, informal caregiving research should extend itself beyond caregiver burden or psychosocial interventions to a wider understanding of social determinants of caregiving.
- **Assess neurocognitive impairments and decision-making capacity during the caregiving course.** Informal caregivers need to be informed about possible

changes in cognition and behaviour to adequately respond to these challenges in their relationships.

- **Develop a positive culture around care and dying.** Hospice culture has a long tradition in some countries but not so in Switzerland**.** A culture of care includes low threshold forms of support (e.g., volunteers) and other forms of social support. Compassionate and caring communities forming a wider care network can support informal caregivers in various ways.
- **Examine policies relating to access to end-of-life options for people with ALS.** This has to be understood as a wider push towards the importance of disability rights and autonomous decision-making at the end-of-life. Especially the non-existence of widespread care homes for invasive ventilation might hinder autonomy of people with ALS to decide according to their own wishes.
- **Examine policies relating to access to assistive technology.** In the future, brain-computer interfaces might be widely available and people with ALS needs opportunities to access this technology.
- **Recognise the work and redistribute financial and welfare support.** While it is important to acknowledge and recognise the work informal caregivers do, it is also important to redistribute resources to enable informal caregivers to provide care. Pressing needs are respite opportunities and less administrative demands on informal caregivers.

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Appendices

Appendix A. Ethics approval

EKNZ

Ethikkommission
Nordwest- und
Zentralschweiz

Präsident
Prof. Christoph Beglinger
Vizepräsidenten
Dr. Angela Frotzler
Dr. Marco Schärer

Herr
Dr. Ch. Poppe
Universität Basel, Insitute for Biomedical Ethics
Bernoullistrasse 28
4056 Basel

Basel, 6. August 2018 / CB

Stellungnahme gemäss HFG Art.51
BASEC Nr Req-2018-00629: Palliative care at home: A multi perspective study on the complex needs of ALS family caregivers

Sehr geehrter Herr Dr. Poppe

Besten Dank für die Einreichung der Unterlagen zur obengenannten Studie datiert vom 30.07.2018 samt Beilagen.

Unsere Abklärungen haben ergeben, dass es sich um keine bewilligungspflichtige Studie im Sinne der kantonalen und eidgenössischen Gesetzgebung handelt, da Ihr Projekt nicht als Forschungsprojekt gemäss HumanForschungsGesetz Art. 2 definiert wird. Aus diesem Grund kann die EKNZ keine formelle Bewilligung ausstellen.

Nach Überprüfung der Anfrage kann die EKNZ jedoch feststellen, dass diese Studie den allgemeinen ethischen Grundsätzen für die Forschung am Menschen entspricht (vgl. Art. 51 Abs. 2 Humanforschungsgesetz).

Ich hoffe, Ihnen mit diesen Angaben zu dienen und verbleibe

mit freundlichen Grüssen

i.V.
Prof. Christoph Beglinger
Präsident der Ethikkommission
Nordwest- und Zentralschweiz / EKNZ

Begutachtete Unterlagen:
- jurisdictional-inquiry-als-care.pdf
- interviewguides-current-former-fokus.pdf

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Appendix B. Interview guides (English translation)

Interview guide former caregivers

Relationship and care situation

1. What was your relationship to the deceased?
2. Can you tell me what the care situation was like and how it evolved?

Support in the course of the disease

3. What support did you have after the diagnosis?
    How could you have been better supported?
    What information would you have needed after the diagnosis?
4. At what point in the illness was the need for support great?
    Where would you have needed more support?
5. How has the care changed over the course of the illness?

Assistance in making changes in daily life

6. What support did you have for caring for the sick person?
    In organizing finances or work?
    To initiate the necessary structural changes?
    Organize the necessary equipment and care material?
    Practical things?
    In case you needed time for yourself or for errands?
    What support would you have wished for?
7. Did you receive support from Spitex?
    How was the cooperation with Spitex?

Assistance with advance care planning

Did your loved one own or have you made decisions about medical treatment in advance?
    What was the advance planning?
    Has an authorized representative or living will been specified?
    How did the advance planning go?
    Who supported you?
    In retrospect, how would you rate the advance planning and support?

End-of-life support for the sick person

9. What support did you need at the end of the life of the sick person?
10. Were you cared for by a mobile palliative care team?
    If so: How was the collaboration with the mobile palliative care teams?

Care for the caregiver support

11. Did you have support with your own worries and needs?
    What helped you?
12. If you are religious, has anyone given you pastoral care?
    If so, how did the pastoral support help you?
13. How were you supported in case of illness or physical discomfort?

Open questions

14. What would have helped you to help the deceased person?
15. What kind of support did you appreciate the most?
16. What would you recommend to best support the patient and family members?
17. Would you like to add something else?

Many thanks.

Interview guide current caregivers

Relationship and care situation

1. How do you relate to the sick person?
2. Can you tell me what the care situation is like at the moment?
3. How has the care situation changed so far?

Support in the course of the disease

4. What support did you have after the diagnosis?
    How could you have been better supported?
    What information would you have needed after the diagnosis?

5. At which point of the illness or in which situation was the need for support great?
 Where would you have needed more support?

Assistance in making changes in daily life

6. What support do you have for caring for the sick person?
 In organizing finances or work?
 To initiate the necessary structural changes?
 Organize the necessary equipment and care material?
 Practical things?
 If you need time for yourself or for errands?
☐ If not, would that be helpful?
7. Do you have support from Spitex?
 How is the cooperation with Spitex?

Assistance with advance care planning

1. Did your loved one own or have you made decisions about future medical treatments?
    What is advance planning?
    Has an authorized representative or living will been specified?
    How did the advance planning go?
    Who supported you?
    Who could support you?
    How would you rate the support?

End-of-life support for the sick person

8. What support do you need at the end of the life of the sick person?
9. Are you currently being looked after by a mobile palliative care team?
 If so: How is the collaboration with the mobile palliative care team?

Care for the caregiver support

10. Do you have support with your own worries and needs?
    What helps you?
11. If you are religious, does someone provide pastoral support?
    If so: How does the pastoral support help you?
12. How are you supported in the event of your own illness or physical discomfort?

Open questions

1. What helps you to help the sick person?
2. What kind of support do you value most?
3. What would you recommend to best support the patient and family members?
4. Would you like to add something else?

Many Thanks.

Interview guide healthcare professionals

Introductory questions

How long have you been working with people with ALS and their families?
What are your main tasks in the area of ALS?
At what stage do you have contact with ALS patients and their families? For example, directly after the diagnosis, at the end of life ...
Did you complete specific training that prepared you for the care of ALS patients and their families?

Difficult situations

How do you perceive your work with people with ALS and their families?
Can you tell me about a situation that was particularly difficult for someone with ALS and their family and in which you were able to support the family?

Informal caregivers‘ need for support

What general need for support do you see in relatives who care for someone with ALS?
What kind of support options, e.g. religious, practical, financial or other, do you think are important for relatives?
Which are the most important in your professional experience?
According to your professional experience, which ones are missing?
Specific support: advance planning, palliative care, training ...
In which special areas do you support patients and their relatives? Examples: advance care planning, palliative care, training of skills?
Can you tell me about any challenges in this area?

After bereavement

What contact do you have with families after the patient's death?
What support do relatives need during the grief phase?
collaboration
How would you describe the employment relationship between you and your family caregivers?
What are the challenges for non-specialized nurses when dealing with ALS patients and their families?
What should health professionals be able to or offer to support caregiving relatives?

Own support needs

What support needs do you have yourself in your work with ALS patients and relatives?
Is there something you want to discuss on the subject that I didn't ask?
Thank you for the interview!

Appendix C. PROSPERO Protocol

PROSPERO

International prospective register of systematic reviews

Citation

Christopher Poppe, Insa Koné, Luzia Iseli, Bernice Simone Elger, Tenzin Wangmo. Differentiating needs of informal caregivers of ALS patients across the caregiving course: a systematic review. PROSPERO 2018 CRD42018105311 Available from:
https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42018105311

Review question

What are the needs of informal caregivers of patients with ALS across caregiving course? How do needs vary over different stages of caregiving?

Searches

From 1. August to 1. October 2018, these databases will be searched: EMBASE, MEDLINE (PubMed), PsycINFO, CINAHL.

We will search for variations of the following terms:

Population: Informal/family caregivers

Context: Amyotrophic Lateral Sclerosis/Motor Neuron Disease

Outcome: Needs

We include literature from 2000 to 2018. We chose this time frame to update the last comprehensive review, which included literature up to 2011, and to guarantee a sufficient amount of studies. We include publications
in both English and German and adapt the search terms respectively.

After consultation with a librarian, the search was restricted to population and context.

Other sources of references include: Citation tracking, reading references, and extracting references from reviews.

Types of study to be included

Inclusion:

(1) Empirical studies with quantitative, qualitative, and mixed-method study designs; AND

(2) Study participants of the included studies should be informal caregivers of ALS patients and/or ALS patients themselves. This means that we consider both the perspective of caregivers as well as the perspective of patients on caregivers' needs; AND

(3) Describing the needs of informal caregivers or should be distinguishable.

Exclusion: Comparative studies of caregivers of different patient groups where needs for ALS caregivers cannot be extracted separately, reviews, commentaries, letters to the editor, editorials and unpublished works

Condition or domain being studied

Informal caregivers, especially family members, provide the primary care for patients with amyotrophic lateral
sclerosis (ALS), or motor neuron disease (MND). ALS is a life-limiting neurodegenerative disease with a vast
array of symptoms, especially progressive muscle atrophy, fasciculations, dysphagia, and dyspnoea. The median life expectancy after first onset of symptoms is 3 years.

The domain studied in this review is informal caregiving for patients with ALS across the caregiving course from first onset of symptoms to bereavement.

We assume that stages of caregiving are innately linked to disease course; as the disease progresses the caregivers need to adapt. We include the time from first onset of symptoms to diagnosis as stage 0, as there is evidence that the journey to diagnosis significantly impacts caregivers as well as patients. Stages of caregiving after diagnosis are based on the literature: 1. Early coping and adjustment after diagnosis, 2. maintenance, 3. transition to end stage, and 4. coping with change and loss.

Participants/population

Informal/family caregivers of patients with ALS/MND. We define informal caregiving as primary unpaid caregiving for the patient (excluding short term caregivers such as for respite care).

Intervention(s), exposure(s)

The focus of this review is on the multifaceted needs of informal caregivers of patients with ALS/MND at different stages of caregiving, e.g. informational needs at time of diagnosis or supportive needs at the end of life.

Comparator(s)/control

Not applicable.

Main outcome(s)

Multifaceted needs of informal caregivers of patients with ALS across stages of caregiving.

Measures of effect

Not applicable.

Additional outcome(s)

None

Measures of effect

Not applicable.

Data extraction (selection and coding)

At least two reviewers (CP and IK) will screen the retrieved titles and abstracts. If a decision cannot be made upon these, the full text will be accessed for eligibility. If there are discrepancies in judgement, a third reviewer (TW) will be consulted.

For eligible studies, all data will be extracted using a data extraction form in Microsoft Excel. Three reviewers (CP, LI and TW) will extract the following data: study design (qualitative/quantitative/mixed-methods), inclusion and exclusion criteria of participants, year, country, rationale, number of study participants, demographic data, setting, attrition rate, study quality (see below), relevant caregiving stage(s) included, and key outcome(s). If there is more than one stage of caregiving included, key outcomes will be linked to the relevant stage, if possible.

Key outcomes are:

1) From qualitative studies: Themes or descriptions of needs of informal caregivers in self-report or in report of others.

(2) From quantitative studies: needs as outcome on certain scales: Carer Support Needs Assessment Tool (CSNAT), Caregiver Needs Assessment (CNA), other instruments for caregiver needs assessment. All data is linked to a stage of caregiving. If these formal assessment tools are not used, needs arising from quantitative studies will be described narratively.

Risk of bias (quality) assessment

For qualitative studies, quality is assessed using a checklist. Two authors (CP and TW) will independently assess the quality of qualitative studies by using the Joanna-Briggs-Institute (JBI) checklist for qualitative research, a well-researched tool for the evaluation of qualitative studies. Disagreements between the two authors (CP and TW) over the risk of bias in particular studies will be resolved by discussion, with involvement of a third reviewer when necessary.

For quantitative studies, sample size and comprehensive report of statistical data are the primary quality indicators.

For these quantitative criteria, we have opted for an adapted version of the NIH Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies checklist.

Risk of bias is assessed in accordance with the Cochrane Handbook chapter 8 . We assume that most studies on the needs of caregivers are non-randomised studies, therefore selection bias (method of randomisation and allocation con-cealment) and performance bias (the use of blinding) are not applicable. In longitudinal study designs, we will check for attrition bias (drop-out of study participants). We will check whether the studies assessed reporting bias.

Mixed-method studies: a combination of the above.

Strategy for data synthesis

Data will be combined by narrative synthesis as it is most suited to combine different research designs and thus comprehensively inform policy. This narrative synthesis will be structured around the different stages of caregiving.

First, we will do a preliminary synthesis by tabulation: the extracted studies will be grouped by research

methodology (qualitative and quantitative; mixed-method data will be split apart by qualitative and quantitative information) in a table. Then, data will be sorted to the different stages of caregiving (0-4). We
will not pool statistical data or conduct a meta-analysis.
Within each caregiving stage subsection, caregivers' needs will be sorted hierarchically by their aggregate ('vote-counting' in narrative synthesis). This hierarchy will influence the importance of the finding in the narrative synthesis.
In a second step, both qualitative and quantitative studies will be analysed by narrative syntheses.
Robustness of synthesis is achieved by taking into account the quality of the studies and providing audit trails for the narrative syntheses.

Analysis of subgroups or subsets

If available, data from different settings (e.g. home care, hospice) will be explored separately.

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Assistant/Associate Professor Tenzin Wangmo. Institute for Biomedical Ethics, University of Basel

Type and method of review

Narrative synthesis, Systematic review

Anticipated or actual start date

01 August 2018

Anticipated completion date

30 April 2019

Funding sources/sponsors

This systematic review is part of a project on informal caregivers of patients with ALS in Switzerland funded by the Swiss Academy of Medical Sciences (SAMS).

Conflicts of interest

Language

(there is not an English language summary)

Country

Switzerland

Stage of review

Review Completed not published

Subject index terms status

Subject indexing assigned by CRD

Subject index terms

Amyotrophic Lateral Sclerosis; Amyotrophic Lateral Sclerosis 2, Juvenile; Caregivers; Humans

Date of registration in PROSPERO

14 August 2018

Date of first submission

27 July 2018

Stage of review at time of this submission

Stage Started Completed
Preliminary searches Yes Yes
Piloting of the study selection process Yes Yes
Formal screening of search results against eligibility criteria Yes Yes
Data extraction Yes Yes

Risk of bias (quality) assessment Yes Yes

Data analysis Yes Yes

Revision note

Status update.

The record owner confirms that the information they have supplied for this submission is accurate and complete and they understand that deliberate provision of inaccurate information or omission of data may be
construed as scientific misconduct.

The record owner confirms that they will update the status of the review when it is completed and will add publication details in due course.

Versions

14 August 2018

Appendix D. Systematic review extraction sheet

A. General Information

1. Paper_ID (Surname first author and year of publication, e.g. Smith2001)

2. Other publications regarding the same study (if applicable, name Paper_IDs of these publications)

3. Date of data extraction

4. Name of person extracting data

5. Title of paper

6. Journal name

7. Year of publication

B. Eligibility

1. Exclusion Criteria (Please tick the most relevant criteria)

   1= Not English or German

   2= No mention of ALS caregivers's needs/not distinguishable

   3= ALS caregivers's needs are not reported by themselves or by the ALS patients (e.g. by healthcare professionals)

2. Publication to be included in full text extraction, if no exclusion criteria is relevant and empiric data collection and analysis is reported:

   1= Yes

   2= No

   Notes:

Only proceed with specific data extraction if full text is included in full text extraction!

Qualitative study appraisal (JBI)	Yes	No	Unclear	Not applicable
1. Is there congruity between the stated philosophical perspective and the research methodology?	☐	☐	☐	☐
2. Is there congruity between the research methodology and the research question or objectives?	☐	☐	☐	☐
3. Is there congruity between the research methodology and the methods used to collect data?	☐	☐	☐	☐
4. Is there congruity between the research methodology and the representation and analysis of data?	☐	☐	☐	☐
5. Is there congruity between the research methodology and the interpretation of results?	☐	☐	☐	☐
6. Is there a statement locating the researcher culturally or theoretically?	☐	☐	☐	☐
7. Is the influence of the researcher on the research, and vice-versa, addressed?	☐	☐	☐	☐
8. Are participants, and their voices, adequately represented?	☐	☐	☐	☐
9. Is the research ethical according to current criteria or, for recent studies, and is there evidence of ethical approval by an appropriate body?	☐	☐	☐	☐
10. Do the conclusions drawn in the research report flow from the analysis, or interpretation, of the data?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

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Quantitative study appraisal (NIH)

	Yes	No	N/A
1. Was the research question or objective in this paper clearly stated?	☐	☐	☐
2. Was the study population clearly specified and defined?	☐	☐	☐
3. Was the participation rate of eligible persons at least 50%?	☐	☐	☐
4. Were all the subjects selected or recruited from the same or similar populations (including the same time period)? Were inclusion and exclusion criteria for being in the study prespecified and applied uniformly to all participants?	☐	☐	☐
5. Was a sample size justification, power description, or variance and effect estimates provided?	☐	☐	☐
6. Were the outcome measures (dependent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?	☐	☐	☐
7. Was loss to follow-up after baseline 20% or less?	☐	☐	☐

6.-10.,12., 14. deleted

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

C. Specific data extraction

1. Setting

1.1 Country (e.g. Switzerland, Germany, UK, USA)

1.2 Patient setting

1= Inpatient, hospital

2= Inpatient, hospice

3= Outpatient, home

4= Not reported/not applicable

Notes:

2. Aim of study (Please describe in one sentence the aim of the study/research question as stated at the end of the introduction.)

3. Outcome measures (Which primary and secondary outcome measures were described in the methods section? For qualitative methods, 'themes', 'phenomenological description' etc may apply)

4. Methods

4.1 Study design

1= Cross-sectional

2= Longitudinal

3= Experimental (e.g. c-RCT)

4= Other (Specify):

5 = Not reported

Note (describe study design in one sentence as stated by the authors):

4.2 Methodology

1= Qualitative

2= Quantitative

3= Mixed-methods

7= Other (specify):

4.3 Type of data collected

1= Interviews

2= Focus group discussion

3= Survey

4= Medical records

5= Participant observation

6= Register data

7= Combination/Other (Specify): Notes:

4.4 Type of analysis

1= Qualitative – unspecified

2= Qualitative – thematic/content analysis

3= Qualitative – grounded theory/phenomenological analysis

4= Quantitative – descriptive

7= Combination/Others (Specify):

9= Not reported

5. Study population

5.1 Participants

1 = ALS former informal caregivers

2 = ALS current informal caregivers

3 = ALS patients

4 = both ALS current informal caregivers and ALS patients

5.2 Describe study sample in one sentence (e.g. age, health status, gender):

5.3 Planned sample size

N=

Note:

5.4 Actual sample size (report absolute values) N=

Note:

6. Caregiving stage included (tick all that apply)

0. = journey to diagnosis, ie caregivers' needs before a diagnosis is given
1. = early coping and adjustment after diagnosis, ie needs directly after diagnosis
2. = maintenance, ie everything in between adaption after diagnosis and transition to palliative care/end stage
3. = transition to end stage/end stage, ie needs surrounding the transition to the death of the patient, mainly dealing with the loss of respiratory function
4. = coping with change and loss after the death of the patient, ie. needs after death of the patient, prolonged grief

7. Important results linked to the caregiving stages (max. 4 bullet points for qualitative studies; described data for primary and secondary outcome measure for quantitative studies). Results should be sorted into the categories: 1 = Informational, 2 = Practical, 3 = Emotional, 4 = Physical (well-being), 5 = Psychological, 6 = Social, 7 = Spiritual. *Example: 3. 2,5,7 – access to palliative care services, – preparing for the patient's death, – loss of hope*

Notes:

8. Key conclusion of study authors (Please describe max. three bullet points the authors' conclusion.)

9. References of other relevant studies (screen the reference list for any relevant publication)