

## RESEARCH ARTICLE

# Burden and benefit—A mixed methods study of informal Amyotrophic Lateral Sclerosis caregivers in Ireland and the Netherlands

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## Abstract

**Objectives:** Amyotrophic Lateral Sclerosis (ALS) is a systemic and terminal disorder of the central nervous system which causes paralysis of limbs, respiratory and bulbar muscles, impacting on physical, communication, cognitive and behavioural functioning. Informal caregivers play a key role in the care of people with ALS. This study aimed to explore experiences of burden along with any beneficial aspects of caregiving in ALS. An understanding of both burden and benefit is important to support the informal caregiver and the person with ALS.

**Methods/Design:** This exploratory mixed methods study characterizes two groups of informal caregivers in Ireland ( $n = 76$ ) and the Netherlands ( $n = 58$ ). In a semi-structured interview, quantitative data were collected in the form of standardized measures assessing psychological distress, quality of life and burden. Qualitative data were collected from an open ended question, in which caregivers identified positive aspects in their caregiving experience. These data types were purposefully mixed in the analysis and interpretation stages, to provide a greater depth of evidence through diverse research lenses.

**Results:** The caregiver cohorts were predominantly female (69%) and spouse/partners (84%) of the person with ALS. Greater levels of self-assessed burden were found among the caregivers in the Netherlands ( $p < 0.05$ ), and higher levels of quality of life among the cohort from Ireland ( $p < 0.05$ ). Themes generated through qualitative analysis identified caregiver satisfaction, ability to meet the patient's needs and the (re) evaluation of meaning and existential aspects of life as positive aspects of caregiving. Existential factors were identified frequently by the caregivers in Ireland, and personal satisfaction and meeting their care recipient's needs by caregivers in the Netherlands. Three percent of all respondents reported there was nothing positive about caregiving.

**Conclusions:** Based on our findings, we suggest that both burden and the presence of positive factors should be evaluated and monitored. The possibility of concurrent

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positive and challenging experiences should be considered in the design and delivery of supportive interventions for informal caregivers.

#### KEYWORDS

Amyotrophic Lateral Sclerosis (ALS), burden, emotion, informal caregivers, mixed methods, Motor Neurone Disease (MND), positive

#### Key points

- Characterisation of informal caregivers attached to two specialist Amyotrophic Lateral Sclerosis (ALS) multidisciplinary clinics in Dublin and Utrecht.
- Mixed methods analysis articulates the complexity of the caregiving experiences in greater detail than monomethod.
- Identification and description of high burden, psychological distress and quality of life in two caregiver cohorts.
- Evaluation of meaning in life, existential issues and functional, task-oriented elements were among the positive aspects identified.

## 1 | INTRODUCTION

Amyotrophic Lateral Sclerosis (ALS), a sub-type of Motor Neurone Disease (MND), is a systemic and terminal disorder of the central nervous system which causes paralysis of limbs, respiratory and bulbar muscles, cognitive and behavioural decline, and may be accompanied by frontotemporal dementia.<sup>1</sup> There is currently limited treatment, and most people affected by the condition die within 3 years from the onset of symptoms.<sup>2,3</sup> Management of ALS is palliative and aimed at maximising quality of life and minimising the burden of disease for people with ALS and their caregivers.<sup>4</sup> People with ALS are primarily looked after in their own home by informal caregivers, usually family or friends. Informal caregivers are an important component in the ALS care provision system,<sup>5</sup> enabling people with ALS to remain at home rather than going to a care facility.<sup>6</sup>

Caring for a partner or family member with a progressive neurological illness has been recognised as being a source of burden, psychological distress, and impaired quality of life.<sup>7</sup> Physical, cognitive and behavioural impairments of the person with ALS can contribute substantially to the psychological and physical morbidity of the caregiver and affect caregiver burden in ALS.<sup>8</sup> Caregivers of people with ALS may see their own needs as secondary to those of the people with the condition.<sup>9</sup> There are similar findings in other neurological disorders, such as dementia, with patient symptoms and caregiving satisfaction significantly and directly affecting caregiver burden.<sup>10</sup> Research on caregiving commonly focuses on stress and burden and the negative impacts of providing care.<sup>11</sup>

Caregiver burden is frequently mentioned in the research and health care arenas, and results from the multiple physical, psychological, social, and financial stressors associated with caregiving.<sup>12</sup> ALS is a progressive neurodegenerative disorder with death occurring approximately 3–5 years after symptom onset,<sup>13</sup> and it is

particularly important to explore the positive as well as the negative aspects associated with caregiving to support caregiver well-being. Psychosocial interventions should focus on enhancement of the positive aspects of caring which could reinforce caregivers' well-being, resilience and reduce the impact of stress and burden.<sup>14–17</sup> Positive aspects of caregiving for a family member with a progressive neurological disorder can affect adaptation to bereavement in terms of depression and grief.<sup>18</sup>

The aim of this mixed methods, exploratory analysis was to explore both caregiver burden *and* self-described positive experiences of informal caregivers of people with ALS attending two multidisciplinary clinical centres in Dublin and Utrecht. Each year there are 150 new cases of ALS and 380 people living with the condition in Ireland.<sup>13</sup> At least 80% of all ALS patients within Ireland and the Netherlands, attend the National ALS/MND Clinic in Beaumont Hospital, Dublin<sup>19</sup> and University Medical Centre Utrecht respectively.<sup>20</sup>

## 2 | METHODS

### 2.1 | Design

A cross-sectional mixed methods analyses of quantitative and qualitative data collected as part of a longitudinal European multi-centre study of people with ALS – *A Programme for ALS Care in Europe (ALS-CarE)*,<sup>21</sup> in Dublin (Ireland)<sup>1</sup> and Utrecht (The Netherlands).<sup>2</sup>

### 2.2 | Participants

Primary informal caregivers were identified as such by the person with ALS. Primary informal caregivers are defined as those who

previously or currently provide the majority of support and assistance to a person with ALS/MND at any disease stage, without financial compensation.<sup>22</sup> A consecutive sample of caregivers providing care for people with ALS at different stages along the disease trajectory (incident and prevalent cases) were recruited to a study of their own wellbeing. Thus the participating caregivers from the two ALS centres provide care to people with ALS across the disease trajectory, attending those services.

Informed, written consent was followed by an in-person, semi-structured interview conducted in the caregiver's own home by a member of the research team attached to each medical centre. Participants who provided demographic data, completed at least one wellbeing measure and the open-ended question were included in this analysis (Dublin  $n = 76$ ; Utrecht  $n = 58$ ; See Figure 1).

Ethical approval for the use of both patient and caregiver data was received from Beaumont Hospital Ethics (Medical Research) Committee (REC REF 12/84) and the Medical Ethics Committee of the university Medical Center Utrecht (ethics approval code 15-708; national code NL56609.041.16).

## 2.3 | Measures

An interview schedule was developed in collaboration with ALS researchers and clinicians. Pilot tested semi-structured interviews were conducted in the caregiver's own home at a time convenient for them (2015–2017). Caregiver participants were assured of the

confidentiality of the research process and anonymity of their responses. Demographic and socio-economic details were collected during the interview, and a series of standardised measures assessed caregiver burden, psychological distress, and quality of life. In an open-ended question, caregivers were asked to identify what for them are some good things about caregiving. All standardized questionnaires were available in both English and Dutch, and are commonly used in ALS care research.<sup>23</sup>

### 2.3.1 | Psychological distress

The Hospital Anxiety and Depression Scale (HADS)<sup>24</sup> was used to assess psychological distress. Hospital Anxiety and Depression Scale is a statistically reliable 14-item scale composed of two subscales detecting Depression (HADS-D) and Anxiety (HADS-A). The use of a summed HADS total score (HADS-T) is regarded as an adequate estimate of general psychological distress.<sup>25</sup> A HADS-Total cut-off score of  $\geq 12$  was used to identify caregivers with probable psychological distress.<sup>26</sup>

### 2.3.2 | Quality of life

Quality of life was assessed using the Quality of Life in Life-Threatening Illness – Family carer version 2 (QOLLI-F v2).<sup>27</sup> The QOLLI-F v2 consists of 17 items across seven subscales and a single

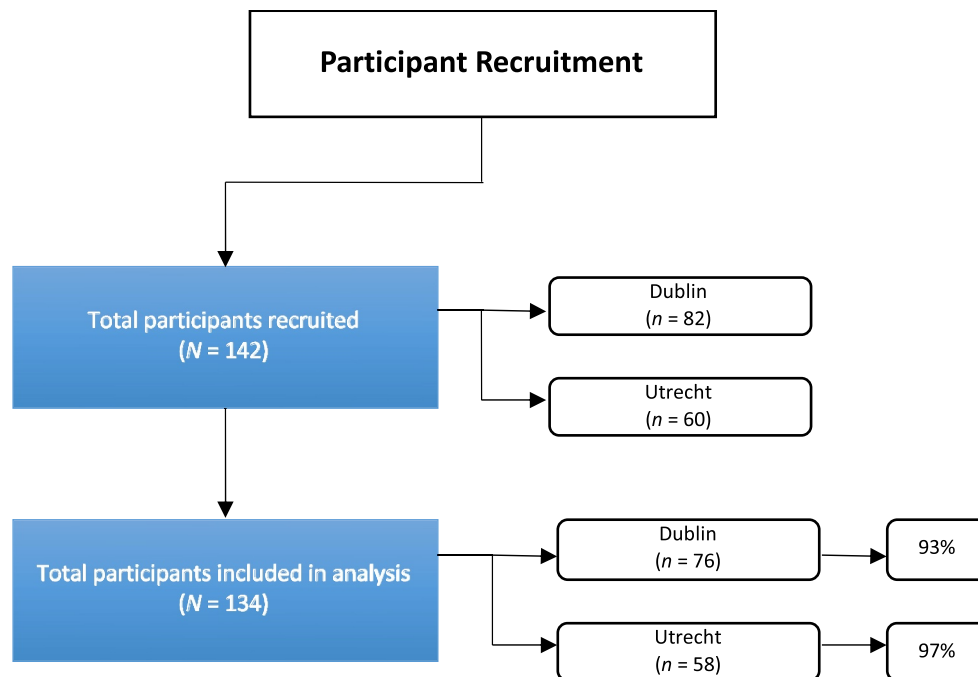


FIGURE 1 Participant Recruitment<sup>†</sup>. † Caregiver participants were included in the analysis if they completed demographic data, at least one measure and the open-ended question

item about overall quality of life. Each item is rated on a 0–10 scale. The Total Score is the sum of the 17 items. In this analysis, the Total Score was used as a measure of self-assessed quality of life, with higher scores indicating greater quality of life.

### 2.3.3 | Caregiver burden

The Zarit Burden Interview (ZBI)<sup>28</sup> is a self-report measure used to assess caregiver burden, with 22 items rated on a 0–4 scale, with a maximum score of 88. A statistically derived cut-off score of  $\geq 24$  indicates burden.<sup>29</sup> Caregivers with a score of  $\geq 24$  were described as being in a 'high-burden' group.<sup>8</sup>

### 2.3.4 | Open-ended question

Qualitative data were collected from a single open-ended question during the interview to gain insight into what caregivers described as positive aspects associated with caregiving and the meanings attached. Caregivers were asked: "For you, what are some things that are good about caregiving?", responses were recorded on paper by the interviewer, and later uploaded into an Excel database. There were no limits placed on the extent of participants' responses. A caregiver could provide a short response or greater detail if they chose to; response density ranged from one word to 147 words.

## 2.4 | Data analysis

### 2.4.1 | Statistical analysis

Descriptive statistics summarized the socio-demographic and well-being measures and are presented as percentage (%), mean (M) and standard deviation (SD) or median (Md) and interquartile range (IQR) as relevant. Normal Gaussian data distribution were tested using the Shapiro-Wilk test and Kolmogorov-Smirnov test. *T*-tests, Mann-Whitney U tests and Chi-square tests compare psychological distress, burden and quality of life among caregivers in Ireland and the Netherlands. Statistical analyses were performed using International Business Machines Corporation (IBM®) Statistical Package for the Social Sciences (SPSS®)<sup>30</sup> version 26.

### 2.4.2 | Qualitative analysis

The 'codebook' approach to thematic analysis was used to identify, analyse and report themes from caregiver responses in a multi-phase process including initial coding, theme development, review and definition.<sup>31</sup> Initial coding was carried out by two coders

(authors EC and PK). An inductive approach was driven by the content of the data, with both descriptive and interpretative approaches used during theme development and refinement.<sup>32</sup> The coding frame was applied to the Irish and Dutch (translated to English) data separately. There was discussion on points of agreement/disagreement leading to consensual validation.<sup>33</sup> The codes generated and themes constructed in this analysis were reviewed and credibility of findings were established based on clinical experience. Microsoft Excel v16.49 (2021) was used to collate and manage the qualitative data, to audit record coding patterns and theme development.

## 3 | RESULTS

### 3.1 | Caregiver characteristics

#### 3.1.1 | Overview

Caregiver characteristics are presented in Table 1 ( $N = 134$ ). Overall, most respondents were female (69%), and lived with the person with ALS (88%). The majority of caregivers had completed post-secondary level education (54%), self-assessed health was excellent/very good/good (85%), while at the same time 38% had a long-term illness or disability.

#### 3.1.2 | Caregivers – Amyotrophic Lateral Sclerosis centre Dublin

The majority of this cohort was female (73.7%), and family caregivers – spouse/partner (78.9%); adult child (13.2%); other family (parent 2.6%, sibling 2.6%). Eighty-eight percent lived with the care recipient. The mean age of caregivers was 57 years, range 27–81 years. They spent an average of 35 h per week providing care ( $Md = 9.5$  h). Eighty-five percent rated their own health as either excellent, very good or good, while 40% said they also had long-term health problems. Thirty seven percent of the cohort completed a degree or higher level of education, and 43% were employed at the time of their interview.

#### 3.1.3 | Caregivers – Amyotrophic Lateral Sclerosis centre Utrecht

This cohort was also predominantly female (61.8%) with over one-third male. They were family caregivers – spouse/partner (89.1%); adult child (3.6%); other family (sibling 3.6%). There were no parental caregivers. Eighty-nine percent lived with the ALS patient for whom they provided care. The average age was 65 years, ranging from 45 to 80 years. These caregivers spent an average of 96.9 h a week

**TABLE 1** Caregiver characteristics  
(*n* = number of participants, SD =  
Standard Deviation, IQR = Interquartile  
Range.)

Characteristics	ALS centre	
	Dublin ( <i>n</i> = 76 <sup>a</sup> )	Utrecht ( <i>n</i> = 58 <sup>a</sup> )
Caregiver age (years)		
Mean (SD)	57.03 (13.75)	65.41 (7.88)
Median (IQR)	58.52 (18.63)	66.05 (9.82)
Range	26.78–80.84	44.74–80.13
Sex <i>n</i> (%)		
Male	20 (26.3%)	21 (38.2%)
Female	56 (73.7%)	34 (61.8%)
Relationship to patient <i>n</i> (%)		
Spouse/partner	60 (78.9%)	49 (89.1%)
Son/daughter	10 (13.2%)	2 (3.6%)
Sibling	2 (2.6%)	2 (3.6%)
Parent	2 (2.6%)	-
Other <sup>b</sup>	2 (2.6%)	2 (3.6%)
Living with patient <i>n</i> (%)		
Yes	66 (88%)	49 (89.1%)
No	9 (12%)	6 (10.9%)
Education level completed <i>n</i> (%)		
Primary school	13 (17.3%)	-
Secondary school	19 (25.4%)	28 (50.9%)
Post-secondary/Technical/Apprenticeship	15 (20%)	19 (34.5%)
Degree or higher	28 (37.3%)	8 (14.5%)
Current employment status <i>n</i> (%)		
Employed	31 (43.1%)	18 (32.7%)
Retired	24 (33.3%)	27 (49.1%)
Hours of care provided to patient (per week)		
Mean (SD)	35.11 (54.16)	96.86 (73.65)
Median (IQR)	9.5 (38.75)	142.5 (150.75)
Range	0–168	1–168
Health status <i>n</i> (%)		
Excellent/very good/good	63 (85.1%)	47 (85.4%)
Fair	8 (10.8%)	7 (12.7%)
Poor	3 (4.1%)	1 (1.8%)
Long term illness, health problems or disability <i>n</i> (%)		
Yes	24 (41.4%)	19 (34.5%)
No	34 (58.6%)	36 (65.5%)

<sup>a</sup>Some caregivers did not complete all questions and measures.

<sup>b</sup>Includes aunt, brother-in-law, stepson and neighbours.

providing care (*Md* = 142.5 h). Eighty-five percent self-rated their own health as excellent, very good or good, while 34.5% said they also had long-term health problems. Fifteen percent of the Dutch

cohort completed a degree or higher level of education, and approximately 33% were in employment at the time of their interview.

Outcome measures	ALS centre		Sig. value
	Dublin (n = 76 <sup>a</sup> )	Utrecht (n = 58 <sup>a</sup> )	
Burden	n = 71	n = 53	
ZBI Total, M (SD)	14.7 (11.28)	23.17 (13.57)	p = 0.0002 <sup>d</sup>
High burden <sup>b</sup> , n (%)	15 (21.1%)	26 (49.1%)	p = 0.004 <sup>d</sup>
Low burden <sup>b</sup> , n (%)	56 (78.9%)	27 (50.9%)	
Psychological distress	n = 72	n = 54	
HADS-Total, M (SD)	12.74 (8.59)	10.76 (6.91)	p = 0.306
Probable Distress <sup>c</sup> , n (%)	31 (43.1%)	21 (38.9%)	p = 0.774
HADS-Anxiety, M (SD)	7.86 (4.65)	5.85 (3.4)	p = 0.018 <sup>d</sup>
HADS-depression, M (SD)	4.88 (4.41)	4.91 (4.05)	p = 0.686
Quality of life	n = 69	n = 45	
QOLTI-F Total, M (SD)	7.22 (1.38)	6.51 (1.08)	p = 0.004 <sup>d</sup>

<sup>a</sup>Some caregivers did not complete all questions and measures.

<sup>b</sup>Cut-off score of  $\geq 24$  for High Burden.<sup>29</sup>

<sup>c</sup>Cut-off score of  $\geq 12$  for probable psychological distress.<sup>26</sup>

<sup>d</sup>The mean difference is significant at the 0.05 level.

TABLE 2 Caregiver Wellbeing Outcomes (n = number of participants, M = mean, SD = standard deviation)

## 3.2 | Caregiver wellbeing

### 3.2.1 | Overview

The averages of the wellbeing measures for all caregivers were as follows: caregiver burden (M = 18.32, SD = 12.96); psychological distress (HADS-T [M = 11.89, SD = 7.95]; HADS-A [M = 7, SD = 4.26]; HADS-D [M = 4.89, SD = 4.24]), and quality of life (M = 6.94, SD = 1.31). As per scale cut-offs, 29.9% (n = 40) were classified as 'highly burdened', and 38.8% (n = 52) as having 'probable psychological distress'. Wellbeing outcomes for caregivers from each clinical centre are presented in Table 2. A comparison of caregiver burden, psychological distress and quality of life scores between countries are presented in the following paragraphs.

### 3.2.2 | Caregiver burden

Mann Whitney U Tests show that mean levels of burden were significantly higher in the Dutch cohort (Md = 23, IQR = 19.5, n = 53) compared to the cohort from Ireland (Md = 11, IQR = 16, n = 71), U = 2611, z = 3.69, p = 0.0002, r = 0.33. Categorisation into high and low burden (ZBI =  $\geq 24$ ) indicated 21% in the Irish cohort and 49% in the Dutch cohort were highly burdened. A chi-square test (with Yates' correction for continuity) indicated a significant association between national centre and burden categorisation,  $\chi^2$  (1, n = 124) = 8.265, p = 0.004.

### 3.2.3 | Psychological distress

There were no significant differences in psychological distress (HADS-T) scores between the Irish (Md = 10, IQR = 12.75, n = 72) and Dutch caregivers (Md = 9, IQR = 9, n = 54), U = 1736.5, z = -1.024, p = 0.306, r = 0.09. There were no significant differences in the average depression scores (HADS-D) between Irish (Md = 4, IQR = 6.75, n = 72) and Dutch caregivers (Md = 3, IQR = 6, n = 54), U = 2025.5, z = 0.404, p = 0.686, r = 0.04. However, average levels of anxiety (HADS-A) were significantly higher among the Irish cohort (Md = 8, IQR = 7, n = 72) compared to the Dutch cohort (Md = 5.5, IQR = 4.25, n = 54), U = 1467, z = -2.359, p = 0.018, r = -0.21. The magnitude of the difference in the means (mean difference = 2.009, 95% CI: 0.59 to 3.43) was moderate (eta squared = 0.059). A chi-square test for independence (with Yates' Continuity Correction) indicated no significant association between country and probable psychological distress categorisation,  $\chi^2$  (1, n = 126) = 0.083, p = 0.774.

### 3.2.4 | Quality of life

An independent-samples t-test compared the quality of life (QOLTI-F Total) of the caregiver cohorts, and showed a significant difference, with higher quality of life reported by the caregivers in Ireland (M = 7.23, SD = 1.38); (M = 6.51, SD = 1.08); t (112) = 2.907, p = 0.004, two-tailed. The magnitude of the difference in the means (mean difference = 0.708, 95% CI: 0.23 to 1.20) was moderate (eta squared = 0.073).

TABLE 3 Qualitative analysis: Theme frequencies and illustrative quotes (centre and burden level)

Themes & Quotes	ALS centre	
	Dublin	Utrecht
<b>Meaning in Life</b>	47 (43.9%) <sup>a</sup>	18 (27.3%)
<b>Including the importance of time, relationships, existential issues, enjoyment and aspects like closeness, trust etc.</b>		
<i>'It refocuses the mind on what are the really important things in life.'</i> - caregiver, Dublin (low burden)		
<i>'It has connected us.'</i> - caregiver, Utrecht (high burden)		
<i>'Being strong together'</i> - caregiver, Utrecht (low burden)		
<i>'being together, spend more time together.'</i> - caregiver, Dublin (high burden)		
<i>'the fact that you have more intensive contact with each other and talk more with each other.'</i> - caregiver, Utrecht (low burden)		
<i>'We come to a closure of our life together.'</i> - caregiver, Utrecht (high burden)		
<b>Personal Satisfaction</b>	39 (36.4%)	22 (33.3%)
<b>Including satisfaction, being able to provide care, being healthy enough, role, duty, self-development, reciprocity.</b>		
<i>'Thankful for good health to be able to care for them.'</i> - caregiver, Dublin (low burden)		
<i>'My own qualities emerge: Being self-supportive, creative and leadership [...]. No matter how hard it is, it brings out a lot of good in you. Because of this situation I have a lot more understanding of ill people or people who have been through something nasty. My capacity to feel empathetic has grown much bigger. Appreciation and respect for myself because of the care I give'</i> - caregiver, Utrecht (high burden)		
<i>'You have to, it is your role. If I was sick, my wife would do it for me.'</i> - caregiver, Dublin (low burden)		
<i>'to give something back to my parents'</i> - caregiver, Utrecht (high burden)		
<b>Meeting the Patient Needs</b>	6 (5.6%)	19 (28.8%)
<b>Including being able to keep patient at home, caring for patient themselves, patient wellness, giving patient a good quality of life, good health services available, and finding external support helpful.</b>		
<i>'makes them feel good that they have someone close to them rather than strangers.'</i> - caregiver, Dublin (low burden)		
<i>'In this way my wife can stay in her own environment [her home].'</i> - caregiver, Utrecht (high burden)		
<i>'Letting him remain independent as much as he possibly can.'</i> - caregiver, Dublin (low burden)		
<i>'I am glad to be able to do this for him in the last phase of his life in the place where he wants to be and where he can feel safe and where he will keep feeling involved and where he will keep experiencing all that goes on in his family/household.'</i> - caregiver, Utrecht (high burden)		
<i>'the advices and help from the rehabilitation centre'</i> - Caregiver, Utrecht (high burden)		
<b>Nothing Positive</b>	3 (2.8%)	2 (3%)
<i>'nothing good about it. I don't see what is good about it.'</i> - Caregiver, Dublin (low burden)		
<i>'at the moment not yet'</i> - Caregiver, Utrecht (low burden)		
<b>Other</b>	12 (11.2%)	5 (7.6%)
<b>Including 'I don't know', 'Most of it is good', 'Can't say, not doing it', and 'so far so good'.</b>		
<i>'not sure yet'</i> - caregiver, Utrecht (low burden)		
<i>'I don't know how to answer that to be honest. It's getting on with life really isn't it.'</i> - Caregiver, Dublin (low burden)		

<sup>a</sup>The frequency represents the number of responses coded as that theme, as a percentage of all coded responses in that centre.

### 3.3 | Qualitative analysis

“For you, what are some things that are good about caregiving?”

While recognising that accounts and experiences are socially mediated, we took a realist interpretative approach to the analysis of the qualitative data. Following hybrid inductive and deductive coding, theme definition and refinement, four themes and composite sub-themes were generated from the coded responses (See Table 3). A response could be coded to more than one theme.

Some caregivers (2%), responded that the question did not apply to them at the time of the interview while 3% said they could not think of anything good about caregiving. For those respondents who indicated positive factors associated with caregiving, three main themes were generated from the response data: ‘*Meaning in Life*’, ‘*Personal Satisfaction*’ and ‘*Meeting the Patient Needs*’.

There was a variety of responses around meaning and purpose in life, closeness of relationships and the importance of time to spend with a loved one, for example, “*It refocuses the mind on what are the really important things in life.*” (Caregiver – Dublin). Positive aspects identified by caregivers, included feelings of satisfaction and being healthy and able to provide care, for example, “*Thankful for good health to be able to care for them.*” (Caregiver – Dublin). Support received from the health services and meeting the patient needs was seen to be positive factors, for example, “*the advice and help from the rehabilitation centre.*” (Caregiver – Utrecht). The reciprocal relationship and role of spouse/partner was mentioned, for example, “*If I was sick, my wife would do it for me.*” (Caregiver – Dublin).

The frequency of each theme as a percentage of all the themes identified by the respondents in each centre is presented below (Table 3), with illustrative quotes and attributions by national cohort and burden categorisation (high-low).

Figure 2 illustrates the frequency with which the three themes were mentioned by caregivers in the Dublin and Utrecht cohorts. It is noteworthy, that *Meaning in Life* and *Personal Satisfaction* were mentioned more often by caregivers in Dublin compared with those from the Dutch centre. The positivity acknowledged from *Meeting the Patient Needs* was identified more often by caregivers in the Utrecht cohort.

Results from an integrated analysis of the quantitative and qualitative data are presented in Table 4 and Figure 3 below. Here we see the frequency of themes mentioned by caregivers dichotomised by high and low burden classification.

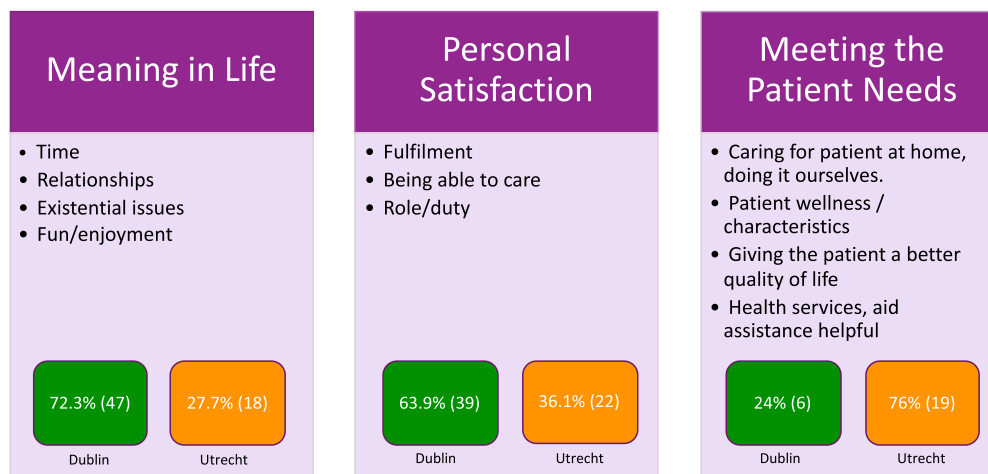
Levels of caregiver burden and thematised beneficial aspects are demonstrated in Table 4, - *Meaning in Life* and *Personal Satisfaction* were mentioned more often by Irish caregivers in the low burden category; *Personal Satisfaction* and *Meeting the Patient Needs* were mentioned more frequently by the high burdened caregivers in the Netherlands.

## 4 | DISCUSSION

Caring for someone with a neurodegenerative disease, such as ALS, is challenging, and can impact negatively on the wellbeing of the caregiver. Nevertheless, when asked to identify some positive as-

### “For you, what are some things that are good about caregiving?”

- Themes, subthemes, and frequency of mentions per ALS Centre.



† Frequency of themes mentioned by caregiver in each ALS centre.

\* “nothing positive” 3% and “other” 10% overall.

FIGURE 2 Coding framework and frequencies<sup>†\*</sup> - positive aspects of caregiving. † Frequency of themes mentioned by caregiver in each ALS centre. \* “nothing positive” 3% and “other” 10% overall.



TABLE 4 Frequency of themes and dichotomised burden level

Theme	Dublin (n = 76 <sup>a</sup> ) n (%) <sup>b</sup>		Utrecht (n = 58 <sup>a</sup> ) n (%) <sup>b</sup>	
	Low burden	High burden	Low burden	High burden
Meaning in life	26 (76.5%)	8 (23.5%)	10 (55.6%)	8 (44.4%)
Personal satisfaction	24 (77.4%)	7 (22.6%)	6 (33.3%)	12 (66.7%)
Meeting the patient needs	6 (85.7%)	1 (14.3%)	6 (37.5%)	10 (62.5%)
Nothing positive	2 (66.7%)	1 (33.3%)	2 (100%)	0 (0%)
Other	10 (83.3%)	2 (16.7%)	4 (80%)	1 (20%)

<sup>a</sup>Not all caregivers completed the ZBI to be categorised into a burden category.

<sup>b</sup>Percent of caregivers in the burden category of that ALS centre per theme where responses could be coded to more than one theme.

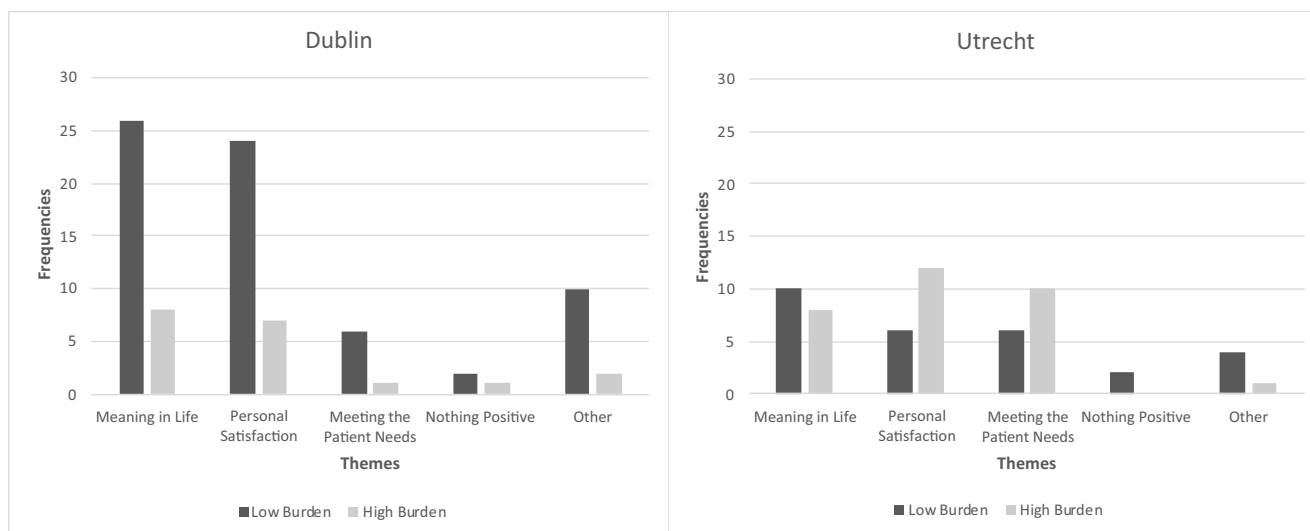


FIGURE 3 Integrated analysis: theme frequency by Amyotrophic Lateral Sclerosis (ALS) centre and burden level (low/high)

pects, respondents described what they considered to be good things related to their caregiving experiences.

In line with previous research, most informal caregivers were female, spouses/partners of the person with ALS and were living with the care recipient. Over one-third of the Dutch cohort was male. There were more adult children among the Irish caregivers. The Dutch were on average 8 years older than their Irish counterparts. Between 30 and 40% of all caregivers were employed; and despite a substantial number indicating long-term illness, self-assessed health ranged from good to excellent for the majority.

Our findings identifying the negative impacts associated with caregiving in ALS, supports previous research,<sup>6,19,34</sup> and acknowledges the enormous challenges encountered. Caregivers are confronted with changing realities, bodily deterioration, increasing levels of dependency, and the changing nature of relationships.<sup>35</sup>

In terms of burden, there was a significant difference in mean burden levels for the two cohorts. When categorised according to high and low burden that is, mean ZBI of greater than or equal to 24,<sup>29</sup> one fifth of caregivers in the Irish group were in the high burden category with approximately half of the Dutch group thus categorised. Looking at psychological distress, higher mean anxiety levels were recorded for the caregivers in Ireland, and 43% were

above the cut-off score for 'probable' psychological distress. Quality of Life scores were lower in the Dutch cohort.

Our mixed methods analysis evidences the co-occurrence of positive and less positive experiences for these caregivers. Although negative outcomes remain an important focus of research, the potential rewarding and positive outcomes of the caregiving experience should be acknowledged.<sup>11</sup> Many of the caregivers in this study experienced a sense of satisfaction as they help somebody they are close to, they felt needed, useful, and good about themselves.<sup>36,37</sup> The positive aspects identified included (re)evaluation of what matters, adding meaning to life, and feeling useful and needed, supporting those found by Baronet.<sup>38</sup> There was a sense of fulfilment for meeting a duty or obligation and enjoyment derived from caregiving itself or from companionship with the care recipient<sup>16</sup> and moments of humour.<sup>39</sup> Like Tarlow et al.,<sup>36</sup> this analysis shows that for many, caregiving enabled them to appreciate life more and strengthened their relationships with loved ones.<sup>37</sup>

Factors thematized under of 'Meaning in Life' were mentioned by almost half of Irish caregivers, and less frequently in the Dutch cohort. Indeed, within this theme, *time to spend together* emerged as an important positive factor for the Irish caregivers but was not mentioned by the Dutch group. Positive aspects related to the theme of

'Meeting Patient Needs' and the availability of good health services were mentioned frequently by the caregivers in the Netherlands. An integrated analysis illustrated the frequency of themes across the high/low burden categories, where 'Personal Satisfaction' and 'Meeting the Patient Needs' were described often by high-burden Dutch caregivers.

Cultural and social factors are related to self-reported symptoms of anxiety and depression among informal caregivers.<sup>40</sup> We can speculate that differential emphasis on spiritual and existential elements, patient impairment (e.g., functional and behavioural) and the national health services arrangements are culturally-specific explanatory variables, however further studies are required to explore these.

The negative and positive aspects of caregiving for someone with ALS are interrelated and affect coping strategies.<sup>41</sup> Benefit-finding is the ability to find meaning through positive reappraisals, spiritual beliefs or other adaptive coping mechanisms in the face of stress.<sup>42</sup> Finding benefit in the caregiving role or tasks can be seen as a positive form of coping with stressful circumstances, and a buffer to the more negative aspects.<sup>16</sup>

The Dutch ALS services are structured according to the diagnostic, rehabilitation, and terminal phases<sup>37</sup> of the disease, with transfer from the hospital based neurology team to a network of rehabilitation teams. In Ireland, ALS care is centralized by a national team that provides ongoing outreach and home visits throughout the patient journey from diagnosis to end of life. The commonalities and differences in the experiences of two cohorts of informal caregivers point to the importance of understanding individual, cultural and system-level characteristics. Support needs of caregivers differ within and between cohorts, and it is important to provide socially and culturally informed care management.<sup>37</sup>

#### 4.1 | Strengths and limitations

The caregivers in this study were care partners of people with ALS who attended specialist clinics in Dublin and Utrecht respectively. We do not claim that our findings generalise to other ALS informal caregivers.

It is important to identify the characteristics of caregivers, the negative impacts of their role and concurrent positive aspects of informal care provision in order to design specific interventions and supports. As ALS is a rare disease, it is crucial to learn from our international counterparts as much as possible to better inform clinical care for supportive interventions and to better understand the complexity of experiences. The cross-country multi-site design and methodology of this study facilitates this.

The combination of quantitative data from standardised measures of burden, psychological distress, and quality of life and qualitative data on the self-identified positive aspects provided unique insights into the caregivers' experiences. Categorising respondents into high/low burden, *Meaning in Life* was mentioned most often by low-burdened caregivers in both cohorts. Whereas

positivity relating to *Personal Satisfaction* and *Meeting the Patient Needs* featured prominently among high-burdened caregivers in the Netherlands.

Factors such as the cognitive and behavioural status of the person with ALS, the stage of disease and burden could be included with a range of covariates in future studies. Additionally, disease-specific factors may impact caregivers' engagement in research, thus the respondents in this study may not be representative of all ALS caregivers. Structural issues such as the configuration of different health services need to be considered, as do culture-specific influences on the expression of distress and burden, and positive things related to caregiving.

Average hours of care provided each week varied across the two study cohorts. The responses of '24/7' were common among the Dutch responses. The Irish caregivers provided more specific details in their responses. A comparison of those figures should be interpreted with caution, pending further study.

Nevertheless, this innovative exploratory study has highlighted the complexity of informal caregiving in this neurodegenerative condition through illustrating the concurrence of burden and some positive aspects as described by caregivers themselves.

Further research by this group, with larger sample sizes will build on the findings from this study to inform intervention design and evaluation to support caregivers. In addition, focus group discussions will explore the issues raised from this analysis in greater detail.

## 5 | CONCLUSION

This exploratory mixed methods analysis illustrates the co-existence of both the negative impacts and positive aspects of informal caregiving in ALS. An awareness of possible cross-cultural factors is crucial for the development of international interventions for informal caregivers and for policy development. We must acknowledge that experiences of both burden and benefit can co-exist in order to understand the complexity of informal caregiving.

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### CONFLICT OF INTEREST

Polly Kennedy reports no conflict of interest. Éilís Conroy reports no conflict of interest. Mark Heverin reports no conflict of interest. Iracema Leroi is associate editor of the International Journal of

Geriatric Psychiatry. Anita Beelen reports no conflict of interest. Leonard van den Berg serves on scientific advisory boards for Orion, Orphazyme, Cytokinetics and Biogen Idec; on the editorial boards of ALS and Frontotemporal Degeneration. Orla Hardiman is Editor in Chief of the ALS and Frontotemporal Degeneration Journal. She has been a consultant for Cytokinetics, Alexion, AveXis, Biogen, and Novartis. Miriam Galvin reports no conflict of interest.

#### AUTHOR CONTRIBUTIONS

Polly Kennedy and Éilís Conroy should be considered joint first author. Polly Kennedy, Éilís Conroy, and Miriam Galvin contributed to the manuscript write up. All authors read and approved the final manuscript.

#### INFORMED CONSENT STATEMENT

Written informed consent was obtained from all subjects involved in the study before study enrolment.

#### DATA AVAILABILITY STATEMENT

The data presented in this study are available upon reasonable individual request from Mark Heverin Research Manager in the Academic Unit of Neurology in Trinity College Dublin.

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#### ENDNOTES

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#### REFERENCES

- Hardiman O, van den Berg LH, Kiernan MC. Clinical diagnosis and management of amyotrophic lateral sclerosis. *Nat Rev Neurol*. 2011;7(11):639-649. <https://doi.org/10.1038/nrneurol.2011.153>
- Rooney J, Byrne S, Heverin M, et al. Survival analysis of Irish amyotrophic lateral sclerosis patients diagnosed from 1995-2010. *PLoS ONE*. 2013;8(9):e74733.
- Elamin M, Phukan J, Bede P, et al. Executive dysfunction is a negative prognostic indicator in patients with ALS without dementia. *Neurology*. 2011;76(14):1263-1269. <https://doi.org/10.1212/WNL.0b013e318214359f>
- Connolly S, Galvin M, Hardiman O. End-of-life management in patients with amyotrophic lateral sclerosis. *Lancet Neurology*. 2015;14(4):435-442.
- Galvin M, Gavin T, Mays I, Heverin M, Hardiman O. Individual quality of life in spousal ALS patient-caregiver dyads. *Health Qual Life Outcomes*. 2020;18(1):1-13.
- de Wit J, Bakker LA, van Groenestijn AC, et al. Caregiver burden in amyotrophic lateral sclerosis: a systematic review. *Palliat Med*. 2018;32(1):231-245.
- O'Brien MR, Whitehead B, Jack BA, Mitchell JD. From symptom onset to a diagnosis of amyotrophic lateral sclerosis/motor neuron disease (ALS/MND): experiences of people with ALS/MND and family carers—a qualitative study. *Amyotroph Lateral Scler*. 2011;12(2):97-104.
- Burke T, Elamin M, Galvin M, Hardiman O, Pender N. Caregiver burden in amyotrophic lateral sclerosis: a cross-sectional investigation of predictors. *J Neurology*. 2015;262(6):1526-1532. <https://doi.org/10.1007/s00415-015-7746-z>
- de Wit J, Schröder CD, El Mecky J, Beelen A, van den Berg LH, Visser-Meily JM. Support needs of caregivers of patients with amyotrophic lateral sclerosis: a qualitative study. *Palliat Support Care*. 2019;17(2):195-201.
- Park M, Choi S, Lee SJ, et al. The roles of unmet needs and formal support in the caregiving satisfaction and caregiving burden of family caregivers for persons with dementia. *Int Psychogeriatr*. 2018;30(4):557-567.
- López J, López-Arrieta J, Crespo M. Factors associated with the positive impact of caring for elderly and dependent relatives. *Archives gerontology geriatrics*. 2005;41(1):81-94.
- Hunt CK. Concepts in caregiver research. *J Nurs Scholarsh*. 2003;35(1):27-32.
- Hardiman O, Lefter S, Galvin M. The experience of amyotrophic lateral sclerosis in Ireland. In: RHB, JEK, DO, eds. *Public Policy in ALS/MND Care*. Palgrave Macmillan; 2021: chap The Experience of Amyotrophic Lateral Sclerosis in Ireland.
- Vatter S, Leroi I. Resilience in people with lewy body disorders and their care partners: association with mental health, relationship satisfaction, and care burden. *Brain Sci*. 2022;12(2):148.
- Pinquart M, Sörensen S. Differences between caregivers and non-caregivers in psychological health and physical health: a meta-analysis. *Psychol Aging*. 2003;18(2):250-267.
- Cohen CA, Colantonio A, Vernich L. Positive aspects of caregiving: rounding out the caregiver experience. *Int J Geriatric Psychiatry*. 2002;17(2):184-188.
- Carbonneau H, Caron C, Desrosiers J. Development of a conceptual framework of positive aspects of caregiving in dementia. *Dementia*. 2010;9(3):327-353.
- Boerner K, Schulz R, Horowitz A. Positive aspects of caregiving and adaptation to bereavement. *Psychol Aging*. 2004;19(4):668-675.
- Galvin M, Gaffney R, Corr B, Mays I, Hardiman O. From first symptoms to diagnosis of amyotrophic lateral sclerosis: perspectives of an Irish informal caregiver cohort—a thematic analysis. *BMJ Open*. 2017;7(3):e014985. <https://doi.org/10.1136/bmjopen-2016-014985.20>
- de Jongh AD, van Eijk RPA, Peters SM, et al. Incidence, prevalence, and geographical clustering of motor neuron disease in The Netherlands. *Neurology*. 2021;96(8):e1227-e1236. <https://doi.org/10.1212/WNL.00000000000011467>
- A Programme for Amyotrophic Lateral Sclerosis Care in Europe (ALS-CarE)*. Updated 8 August 2017. Accessed 22 June, 2021. <https://clinicaltrials.gov/ct2/show/NCT03081338>
- Holkham L, Soundy A. The experience of informal caregivers of patients with motor neurone disease: a thematic synthesis. *Palliat Support Care*. 2017;16(4):487-496.
- Paganoni S, Cudkowicz M, Berry JD. Outcome measures in amyotrophic lateral sclerosis clinical trials. *Clin Investig*. 2014;4(7):605-618.
- Zigmond AS, Snaith RP. The hospital anxiety and depression scale. *Acta Psychiatr Scand*. 1983;67(6):361-370.
- Norton S, Cosco T, Doyle F, Done J, Sacker A. The Hospital Anxiety and Depression Scale: a meta confirmatory factor analysis. *J Psychosomatic Res*. 2013;74(1):74-81.
- Pallant JF, Tennant A. An introduction to the Rasch measurement model: an example using the Hospital Anxiety and Depression Scale (HADS). *Br J Clin Psychol*. 2007;46(1):1-18.
- Cohen R, Leis AM, Kuhl D, Charbonneau C, Ritvo P, Ashbury FD. QOLLTI-F: measuring family carer quality of life. *Palliat Med*. 2006;20(8):755-767.
- Zarit SH, Reever KE, Bach-Peterson J. Relatives of the impaired elderly: correlates of feelings of burden. *Gerontologist*. 1980;20(6):649-655.

29. Schreiner AS, Morimoto T, Arai Y, Zarit S. Assessing family caregiver's mental health using a statistically derived cut-off score for the Zarit Burden Interview. *Aging Ment Health*. 2006;10(2):107-111.
30. IBM SPSS Statistics for Macintosh. IBM Corp.; 2019. Version 26.0
31. Braun V, Clarke V. Reflecting on reflexive thematic analysis. Review. *Qualitative Research in Sport Exercise and Health*. 2019;11(4):589-597. <https://doi.org/10.1080/2159676X.2019.1628806>
32. Braun V, Clarke V. *Thematic Analysis*; 2012.
33. Miles MB, Huberman AM. *Qualitative Data Analysis: An Expanded Sourcebook*. Sage; 1994.
34. Galvin M, Carney S, Corr B, Mays I, Pender N, Hardiman O. Needs of informal caregivers across the caregiving course in amyotrophic lateral sclerosis: a qualitative analysis. *BMJ Open*. 2018;8(1):e018721. <https://doi.org/10.1136/bmjopen-2017-018721>
35. Ray RA, Street AF. Caregiver bodywork: family members' experiences of caring for a person with motor neurone disease. *J Adv Nurs*. 2006;56(1):35-43.
36. Tarlow BJ, Wisniewski SR, Belle SH, Rubert M, Ory MG, Gallagher-Thompson D. Positive aspects of caregiving: contributions of the REACH project to the development of new measures for Alzheimer's caregiving. *Res Aging*. 2004;26(4):429-453.
37. Nichter M. Idioms of distress revisited. *Cult Med Psychiatr*. 2010; 34(2):401-416.
38. Baronet AM. The impact of family relations on caregivers' positive and negative appraisal of their caretaking activities. *Fam Relat*. 2003;52(2):137-142.
39. Williams MT, Donnelly JP, Holmlund T, Battaglia M. ALS: family caregiver needs and quality of life. *Amyotroph Lateral Scler*. 2008;9(5):279-286. <https://doi.org/10.1080/17482960801934148>
40. Wulff J, Fänge AM, Lethin C, Chiatti C. Self-reported symptoms of depression and anxiety among informal caregivers of persons with dementia: a cross-sectional comparative study between Sweden and Italy. *BMC Health Serv Res*. 2020;20(1):1-15.
41. Weisser FB, Bristowe K, Jackson D. Experiences of burden, needs, rewards and resilience in family caregivers of people living with Motor Neurone Disease/Amyotrophic Lateral Sclerosis: a secondary thematic analysis of qualitative interviews. *Palliat Med*. 2015;29(8):737-745. <https://doi.org/10.1177/0269216315575851>
42. (APA) APA. Positive aspects of caregiving. 2021. Accessed June 2021. <https://www.apa.org/pi/about/publications/caregivers/faq/positive-aspects>

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