



# BMJ Open Advance care planning in amyotrophic lateral sclerosis (ALS): study protocol for a qualitative longitudinal study with persons with ALS and their family carers

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## ABSTRACT

**Introduction** Amyotrophic lateral sclerosis (ALS) is an incurable motor neuron degenerative disease that has rapid progression and is associated with cognitive impairment. For people with ALS (pALS) and their family carers, advance care planning (ACP) is beneficial, as it can lead to feelings of control/relief and refusal of unwanted treatments. However, evidence concerning the experiences and preferences regarding ACP of pALS and their family carers, especially when their symptoms progress, is scarce. This article describes the protocol for a qualitative longitudinal study that aims to explore: (1) the experiences with ACP and the preferences for future care and treatment of pALS and their family carers and (2) how these experiences and preferences change over time.

**Methods and analysis** A qualitative, longitudinal, multiperspective design. A total of eight to nine dyads (pALS and their family carers) will be recruited, and semistructured interviews administered every 3 months over a 9-month period. Qualitative longitudinal analysis involves content analysis via in-depth reading, followed by a two-step timeline method to describe changes in experiences and preferences within and across participants.

**Ethics and dissemination** This protocol has been approved by the central ethical committee of the University Hospital of Brussels, and local ethical committees of the other participating hospitals (B.U.N. B1432020000128). The results will be disseminated via the research group's ([endoflifecare.be](http://endoflifecare.be)) website, social media and newsletter and via presentations at national and international scientific conferences.

## INTRODUCTION

Amyotrophic lateral sclerosis (ALS) is the most common degenerative motor neuron disease (MND) in adults, affecting the brain and spinal cord.<sup>1</sup> ALS is incurable and characterised by progressive muscle paralysis. Respiratory failure is the most common cause of death.<sup>2,3</sup> The average survival between

## STRENGTHS AND LIMITATIONS OF THIS STUDY

- ⇒ A longitudinal qualitative study design is an ideal method for capturing change in complex processes such as advance care planning (ACP).
- ⇒ Multiperspective interviews will enhance our understanding of the dynamic relationships between people with amyotrophic lateral sclerosis and their family carers.
- ⇒ Monthly telephone calls will help develop a trusting relationship, which might lead to less attrition (attrition is a risk factor in a longitudinal study).
- ⇒ It might be that addressing ACP during the first interviews would trigger the participants to have these conversations, which will be a specific point of attention during the subsequent interviews.
- ⇒ The number of participants is relatively small compared with other longitudinal qualitative studies, but deemed feasible for addressing the aims of this exploratory study.

symptom onset and death is approximately 3 to 4 years, which is significantly shorter than the survival of people with other neurological conditions, such as dementia or multiple sclerosis.<sup>1,4-7</sup> Up to 50% of people with ALS (pALS) also develop a cognitive impairment, such as frontotemporal dementia. Further, they often experience physical, emotional and existential problems that persist until the end of life. However, to date, reports show that the complex needs of pALS often remain unmet.<sup>8-11</sup> Given the incurable nature of ALS, combined with its rapid progression and unmet palliative care needs, an integrated palliative care approach, including advance care planning (ACP), has been widely advocated for this population.<sup>10-12</sup> ACP is defined as a continuous, early-initiated communication process

between patients, their family carers and/or healthcare professionals that enables individuals to define goals and preferences for future end-of-life care. ACP can prepare patients, family carers and healthcare professionals for making the best possible in-the-moment decisions that are consistent with the patients' values, goals and preferences.<sup>13</sup>

A 2014 systematic review in geriatric and cancer populations suggests that ACP can improve communication about goals of care and overall satisfaction with hospital care and end-of-life care,<sup>14</sup> especially if seen as a process with multiple conversations with patients and their family carers occurring over time.<sup>15</sup> However, the majority of studies have investigated ACP practice or participants' perceptions on ACP at one specific timepoint,<sup>16–18</sup> which merely gives a snapshot of the complex and dynamic reality of engaging in the process of ACP throughout the disease trajectory. A 2016 systematic review of ACP in people with MND, including ALS, also showed important benefits with the uptake of ACP—such as feelings of control/relief and refusal of unwanted treatments—but less was found about how and when ACP should be implemented in the care consistent with the persons' and family carers' needs over time.<sup>19</sup>

Most studies on ACP in ALS focus on the risks and benefits of life-sustaining interventions (such as ventilatory support and gastrostomy), identification of a surrogate decision-maker and completing an advance directive.<sup>8,9,20</sup> However, recent recommendations highlight the need for ACP to be seen as a series of broader conversations about hopes, preferences and potential care goals, which can be discussed among patients and family carers themselves in an informal manner.<sup>21,22</sup> Moreover, preferences for current and future care are situational—often related to key events such as symptom progression or multiple hospital admissions—and may change over time.

To our knowledge, only one longitudinal qualitative study about ACP in ALS conducted non-participative observations for 6 months of appointments between pALS and their treating physician, followed by a single in-depth interview with the patients. This study showed the feasibility and acceptability of implementing ACP throughout the pALS disease trajectory.<sup>23</sup> However, only the patients' perspective was considered and not that of their family carers. Up until now, family carers' perspectives about ACP in ALS are mostly explored retrospectively and during bereavement.<sup>24,25</sup> The perspectives of family carers involved in ACP conversations is crucial, as they often have a prominent role in decision-making at the end-of-life,<sup>26</sup> and it has been shown that involvement in ACP improves family carers' confidence when making end-of-life decisions on behalf of their relative (if needed)<sup>27</sup> and reduces the family carers' distress and grieving.<sup>14,28</sup> This shows that serial and multiperspective interviews are ideal in exploring experiences with ACP and preferences for future care and treatments of pALS and their family carers and how these experiences and preferences change over time.

The aim of this article is to describe the protocol of a longitudinal and multiperspective qualitative interview study that aims to explore the experiences of pALS and their family carers with ACP, their preferences for future care and treatments at three different timepoints, and to investigate whether these experiences and preferences change over time. This study will allow us to see in-depth if, how and why ACP occurs and changes in a unique and fast-changing ALS patient population and their family carers.

The research questions are:

1. What are the experiences of pALS and their family carers with engaging in the process of ACP, and how do these experiences change over time?
2. What are the preferences of persons with ALS regarding their future care and treatments, and how do these preferences change over time?
3. What are the preferences of family carers regarding future care and treatments of persons with ALS, and how do these preferences change over time?

In this article, we outline the research design and methodology developed to answer these research questions. ACP is a complex communication process that requires a research design that is capable of exploring such complexity over time. Longitudinal qualitative research is an emerging methodology, in which time is designed into the research process, making change the focus of analysis. Using this methodology, we aim to investigate lived experience of change with regard to ACP and future care and treatment of pALS and their family carers; the processes by which this experience is created; and the causes and consequences of this change. With this protocol, we hope to inform future international longitudinal qualitative research in other populations—such as patients with dementia or organ failure—who could also benefit from receiving optimal ACP delivery, which has so far remained understudied.<sup>29</sup>

## METHODS

### Study design

This study has a qualitative, longitudinal, multiperspective interview design to provide rich information about the ACP process over time from the perspectives of both pALS and their family carers.<sup>30,31</sup> This design is most suitable for exploring an evolving and complex process such as ACP,<sup>32</sup> as this method is driven by a desire to understand, not just if change happens, but how and why it happens in the sociocultural context over time.<sup>32</sup> It offers considerable advantages over more typical 'snapshot' techniques in understanding the participants' changing experiences and preferences.<sup>31</sup> Constructionism will be used as an underlying epistemology,<sup>33</sup> as we want to know more about the views and meanings of pALS and their family carers about experiences with ACP and preferences in future (end-of-life) care in the specific disease trajectory of ALS.

## Setting

This study will follow pALS living in the community and their family carers interviewed at three timepoints on a 3-monthly interval over a period of maximum 9 months. In Belgium, care for pALS is usually organised through neuromuscular reference centres, providing specialist multidisciplinary care comprising expertise in neurology, respiratory care and rehabilitation, as well as in psychology, physiotherapy, occupational therapy, speech and language pathology, nutrition and social work.<sup>34 35</sup> The neuromuscular reference centres are connected to University Hospitals. Care can also be provided by a general practitioner, community-based services (eg, home care nurses) and palliative home care teams. Almost 90% of the patients stay at home even in the terminal stages of the disease.<sup>36</sup> Voluntary support services, in the form of national or regional ALS associations (eg, ALS Liga in Belgium), can also deliver care to pALS and their families.<sup>3</sup>

## Participants and inclusion criteria

We will include patients from three hospitals, all located in Flanders, Belgium, the Northern Dutch-speaking part of Belgium. Inclusion criteria are: (1) the treating neurologist communicated the diagnosis with the pALS and their family carer not more than 6 months ago; (2) pALS and their family carers are older than 18 years; (3) both must sign a written informed consent. Participants will be excluded if they cannot speak in Dutch or if the pALS are diagnosed with frontotemporal dementia. Because the study is an explorative multiperspective study, we decided to include eight to nine dyads of pALS and their family carers to explore if, when and how ACP occurs, which can result in a total of 54 interviews to be analysed (if each participant is interviewed individually three times). This is a smaller sample compared with other longitudinal interview studies.<sup>37 38</sup>

## Recruitment

To recruit pALS and family carers, we have purposefully selected two academic hospitals (UZ Gent and UZ Brussel) and a non-academic hospital (AZ Maria Middelaes). The pALS and their family carers will be approached by their treating neurologist to ascertain willingness to participate in the study. Potential participants who give consent to their neurologist to pass their contact information to the researchers will be contacted by a member of the research team (IV), to address the purpose of the study. We will wait at least 6 weeks after diagnosis to contact the pALS to allow time for grieving after being diagnosed with ALS, which is a life-changing diagnosis.<sup>39</sup>

We will recruit new potential participants when a participating pALS and/or their family carer drops out of the study after the first interview—for example, due to severe deterioration of the illness, death or when they no longer want to participate. Participating in two interviews will give us the opportunity to potentially identify changes in experiences and perspectives (if any). Hence, re-recruitment

is not necessary. We foresee 1 year of recruitment and the data will be collected through face-to-face semistructured interviews with persons with ALS and their family carers in Flanders, Belgium, at three timepoints from February 2021 onwards.

## Data collection

The pALS and their family carers are preferably interviewed separately, as this will give us the opportunity to observe similarities and differences in their experiences and preferences in ACP.<sup>30</sup> However, if the pALS or family carers wish to have the interview together, and both agree, we will honour their request.

Three interview guides have been developed for the first interview: one for the pALS, one for the family carers and one in case the pALS and family carers prefer to be interviewed together. These interview guides focus on: (1) the experience with ALS, (2) the experiences with ACP and (3) the preferences about future care and treatment. The subsequent interviews will build on the previous one and will be adapted based on what has been discussed in the previous interview, to identify possible changes in their experiences and preferences. In the case of a possible change in experiences or preferences, we will reflect during the interview what triggered the change (eg, unexpected hospitalisation), and we will also reflect on whether and how the interviewer had an influence on the possible change (see online supplemental appendix 1).

We aim to interview the participants on a 3-monthly interval, but flexibility in timing is necessary in case of a sudden change in the experiences with ACP and the preferences for future care and treatments. Other studies have shown that it is useful to use telephone contact to assess whether an interview should be brought forward to capture a changing event.<sup>37 40</sup> Therefore, we will conduct short monthly phone calls with the pALS or family carers. During these phone calls, we will ask how the pALS and family carer are doing, how the disease trajectory is evolving and whether a sudden change (eg, unexpected hospitalisation) has occurred. These monthly phone calls are merely a 'check-in' with the pALS or their family carers to assess whether a subsequent interview needs to be planned sooner than anticipated. These phone calls will not be audiotaped or analysed. Prior to the first interview, the interviewer will ask who should be called for the monthly phone calls. If the pALS prefers to be the contact person and speech deteriorates, we will ask whether the family carer may be contacted. These monthly phone calls also help develop trust between participants and interviewer, and they will also help the interviewer monitor possible distress.<sup>29</sup> In case of distress, the interviewer will advise the participants to talk to their treating neurologist or the psychologist of the neurological department. If the pALS's speech deteriorates, and they still wish to participate, they will have the opportunity to participate via a speech computer or in writing.





## Data analysis

Qualitative longitudinal analysis is an iterative and multi-dimensional process, which involves multiple readings of the data. First, content analysis, which involves line-by-line coding, will be used to obtain an in-depth within-case understanding of the data.<sup>41 42</sup> Codes will be constructed in a coding list for each case separately for pALS and family carers and from each interview round. In the case of any discrepancies, the codes will be discussed between two researchers until consensus is reached, which results in a coding list. This coding list will be refined within the research team by grouping the codes into categories and themes.

Second, we will use a two-step timeline method to describe changes in ACP experience and preferences within and over all the participants.<sup>43</sup> First, a timeline—with time on the x-axis and the themes on the y-axis—will be made for each participant of the dyad and each interview to see what has changed over time, how themes will overlap and how they interconnect with each other. Each timeline will give a clear image of the participants' journey and the overarching themes over time. Second, we will use constant comparison within and between the dyads' timelines to delineate characteristic patterns in the sequence of ACP experiences and preferences (both within and between the dyads).<sup>44</sup> To limit subjectivity, results of this timeline method will be discussed within the research team.

## Patient and public involvement

Patients and family carers were involved in the interview guide development. The main results will be disseminated to the study participants. The strategy for the wider dissemination of the study results to pALS and families will be discussed with ALS patient organisations.

## Ethical considerations, ethics approval and dissemination

Ethical approval has been granted by the central ethical committee of the University Hospital of Brussels, (B.U.N. B1432020000128), via an amendment. Ethical approval has been obtained in the other participating hospitals. Given the (possible) vulnerability of pALS and their family carers, various safeguards are considered for this study: (1) serial informed consent will be required in this qualitative longitudinal research. Before the first interview, an informed consent form will be signed. For the other interviews, verbal consent will be obtained by audiotaping. The interviewer will inform the participants of the purpose of the study during the first contact and prior to each interview before audiotaping; (2) the interviewer (IV), who is an experienced clinical psychologist and who is also the main researcher, will take several steps to ensure the participants' comfort prior to and during the interviews (eg, the interview will be conducted at a location and time of the participants' choosing; breaks will be taken throughout the interviews). Given the extensive educational training and experience, she is capable to capture distress if this arises; (3) it will be emphasised

that, if patients or their family carers would decide not to participate, this decision will by no means influence the quality of their care; and (4) participants may deteriorate and die during the study—therefore, it is important that the interviewer him/herself has a supportive network to be well-supported in their role as interviewer.<sup>29</sup>

The results of this study will be submitted for publication in peer-reviewed journals and will be presented at national and international research and professional conferences. Furthermore, we will disseminate the results via the research group's (endoflifecare.be) website, social media and newsletter.

## DISCUSSION

This will be the first study to provide first-hand, longitudinal, in-depth and multiperspective insights into the process of ACP, and this in a unique patient population of ALS and their family carers. Experiences and preferences in ACP may change over time<sup>9 13</sup>—but, so far, no studies have optimally investigated if, how and why ACP experiences and preferences might change over time in pALS. This study will provide highly valuable information for clinical practice concerning when and how to implement ACP throughout the course of the ALS disease, according to the pALS and family carers' views and preferences, which will improve palliative care and end-of-life care in ALS. Moreover, it will also give us insights into how ACP occurs in the informal context, which is currently understudied in the research field. An important strength of the methodology is that a longitudinal qualitative study, if combined with flexibility, is a less restrictive approach towards studying time and change in complex processes such as ACP.<sup>29 45</sup> ACP is usually measured and described in a single point in time, but the disease's complexity cannot be captured via these snapshot techniques. Another important strength is that longitudinal qualitative studies and multiperspective interviews are innovative methods in medicine and especially in the palliative care field. Moreover, longitudinal qualitative research is a prospective approach, but experiences and preferences may change with the perspective of time, which allows us to also have a retrospective view, which requires a unique way of interviewing. A final important strength is that the perspectives of both pALS and their family carers will be interviewed to enhance our understanding of the dynamics and relationships between them and the individual needs of persons with ALS and their family carers in ACP, and this approach will allow us to explore similarities and differences in their views about ACP.

This study has several challenges. First, it is common that participants withdraw in these types of studies because of the longitudinal aspect. We aim to tackle this challenge by having monthly phone calls, as studies have shown that this can make the interviewees feel more comfortable<sup>37 40</sup> and develop a trusting relationship with the interviewer,<sup>29</sup> which could limit participant attrition.<sup>40</sup> Also, if the patient's speech deteriorates, we will give the

pALS the opportunity to tell their story via writing or the use of a speech computer. Nevertheless, a review did show that pALS need some time to work with a speech computer,<sup>46</sup> but before a subsequent interview is planned, we will allow them the sufficient time for working with this speech computer. Another challenge is that recruitment may be difficult since this study does not address cure or treatment. However, previous research has shown that people usually see participating in ACP research as a worthwhile endeavour—and so we consider this challenge to be minor. We will verbally, and with a written informed consent, inform the participants about the purpose of the study in our first contact and before each interview. Discussing ACP is a difficult subject and might be a challenge, especially if the focus is only on end-of-life decisions. However, our focus will be on a broader level of hopes, preferences and potential care goals about the future. Studies have shown that pALS and family carers welcome the opportunity to discuss ACP<sup>9 47</sup> and they regard ACP as something beneficial.<sup>48 49</sup> In this study, we interview pALS and their family carers about if, how and why ACP occurs throughout the disease trajectory. It is possible that, by addressing these topics in the first interviews, the participants will be triggered to think about or discuss ACP, and thus this can influence their views and perceptions on ACP during the subsequent interviews. Therefore, this will be a specific point of attention during the follow-up interviews, and the interviewer will reflect together with the pALS and the family carers on how discussing experiences, assumptions or beliefs about ACP in the previous interviews had an influence on their current experiences with ACP and/or preferences for future (end-of-life) care.

Finally, this study involves a relatively small number of participants (eight to nine dyads) compared with other longitudinal qualitative studies.<sup>37 38</sup> In Belgium, only 220 new ALS diagnoses occur per year,<sup>36</sup> which shows how rare and unique this patient population is. Longitudinal qualitative research inevitably generates a large volume of interviews, for which effective planning is essential to keeping the data manageable. Given the limited timeframe in which to conduct this study, including eight to nine dyads was deemed feasible for addressing the aims of this exploratory study. Hence, we will interpret our results with caution in terms of generalisation to a larger group of pALS and their family carers.

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