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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, multi-perspective design. 8 to 9 dyads (pALS and their family carers) will be recruited for semi-structured interviews every 3 months for 9 months. 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.

Discussion: This will be the first study to better understand the ACP communication process in ALS, which will inform clinical practice on how and when to implement ACP in the care of pALS and their families. The methodology described will inform other researchers on how to conduct longitudinal, multi-perspective qualitative interviews on ACP in other vulnerable patient populations.

Ethics: 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).

Strengths and limitations of this study

- A longitudinal qualitative study design is an ideal method for capturing change in complex processes such as ACP
- Multi-perspective interviews will enhance our understanding of the dynamic relationships between pALS 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 to other longitudinal qualitative studies, but deemed feasible for addressing the aims of this exploratory study

Introduction

Amyotrophic lateral sclerosis (ALS) is the most common degenerative motor neuron disease (MND) in adults, affecting the brain and spinal cord.[1] ALS is incurable and characterized by progressive muscle paralysis. Respiratory failure is the most common cause of death.[2,3] The average survival between symptom onset and death is approximately three to four years, which is significantly shorter than the survival of people with other neurological conditions, such as dementia or multiple sclerosis.[1,4–7] 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.[8–11] 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[10–12]. 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[13].

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[14], especially if seen as a process with multiple conversations with patients and their family carers occurring over time[15]. However, the majority of studies have investigated ACP practice or participants' perceptions on ACP at one specific timepoint,[16–18] 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.[19]

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[8,9,20]. 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[21,22]. 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.[23] 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.[24,25] 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[26], 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)[27] and reduces the family carers' distress and grieving[14,28]. This shows that serial and multi-perspective 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 multi-perspective 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 3 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 people with ALS 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[29].

Methods

Study design

This study has a qualitative, longitudinal, multi-perspective interview design to provide rich information about the ACP process over time from the perspectives of both pALS and their family carers[30,31]. This design is most suitable for exploring an evolving and complex process such as ACP[32], as this method is driven by a desire to understand, not just if change happens, but how and why it happens in the socio-cultural context over time[32]. It offers considerable advantages over more typical 'snapshot' techniques in understanding the participants' changing experiences and preferences[31]. Constructionism will be used as an underlying epistemology,[33] 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 3 timepoints on a 3-monthly interval over a period of maximum 9 months. In Belgium, care for pALS is usually organized through neuromuscular reference centers, 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[34,35]. The neuromuscular reference centers are connected to University Hospitals. Care can also be provided by a general practitioner, community-based services (e.g., home care nurses) and palliative home care teams. Almost 90% of the patients stay at home even in the terminal stages of the disease[36]. Voluntary support services, in the form of national or regional ALS associations (e.g., ALS Liga in Belgium), can also deliver care to pALS and their families[3].

Participants and inclusion criteria

We will include patients from 3 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 multi-perspective study, we decided to include 8 to 9 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 3 times). This is a smaller sample compared to other longitudinal interview studies[37,38].

Recruitment

To recruit pALS and family carers, we have purposefully selected 2 academic hospitals (UZ Gent and UZ Brussel) and a non-academic hospital (AZ Maria Middelares). 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[39].

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 2 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 semi-structured interviews with persons with ALS and their family carers in Flanders, Belgium, at 3 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[30]. 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 upon 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 whether and how the interviewer had an influence on the possible change (see Appendix I).

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[37,40]. Therefore, we will conduct short monthly phone calls with the pALS or family carers. During these phone calls, we will ask how the disease trajectory is evolving and whether a sudden change 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[29]. In case of distress, the interviewer will advise the participants to talk to their physician 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[41,42]. Codes will be constructed in a coding list for each case separately for persons with ALS and family carers and from each interview round. In the case of any discrepancies, the codes will be discussed between 2 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.

Secondly, we will use a 2-step timeline method to describe changes in ACP experience and preferences within and over all the participants[43]. 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)[44]. To limit subjectivity, results of this timeline method will be discussed within the research team.

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 requested 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, will take several steps to ensure the participants' comfort prior to and during the interviews (e.g., the interview will be conducted at a location and time of the participants' choosing; breaks will be taken throughout the interviews); 3) It will be emphasized 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.[29]

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.

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.

Discussion

This will be the first study to provide first-hand, longitudinal, in-depth, and multi-perspective 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[9,13] – 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[29,45]. 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 multi-perspective 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[37,40] and develop a trusting relationship with the interviewer,[29] which could limit participant attrition[40]. 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. 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[9,46] and they regard ACP as something beneficial[47,48]. 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 (8 to 9 dyads) compared to other longitudinal qualitative studies[37,38]. In Belgium, only 220 new ALS diagnoses occur per year[36], 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 8 to 9 dyads was deemed feasible for addressing the aims of this exploratory study. Hence, we will interpret our results with caution in terms of generalization to a larger group of pALS and their family carers.

Conclusion

This study will allow us to see in-depth if, how and why ACP occurs and changes in a unique patient population of ALS and their family carers. This protocol can inform other international researchers on how to conduct longitudinal, multi-perspective qualitative interviews in other populations who could also benefit from receiving ACP, but for whom the evidence concerning how ACP can be delivered optimally remains scarce.

Authors contribution

Conception and design of the work: IV, JDB, EC, AVDH, LVDB, LD, ADV; Ethic approval: IV, JDB, EC, AVDH, LVDB, LD, ADV; Drafting the work: IV; Critical revision for intellectual content: IV, RM, JDB, EC, AVDH, LVDB, LD, ADV; All authors have read and approved the final manuscript.

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Competing interests statement

The authors declare that they have no competing interests.

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Appendix I: interview guides

Interview guide 1: Person with ALS (interview 1)

Introduction

XX months ago you've received the diagnosis of ALS from XX (name neurologist). Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first symptoms/signals?
- When did you notice that something was wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

Can you tell me something more about ALS? What impact it has on you?

- Who told you this?
- What did you think/feel when you heard about this?

Do you think about your future? Is it something you think about it more than before the diagnosis or vice versa? How do you see your future now?

Option 1: Patient says (s)he thinks about the future:

- What are your expectations about the future? What do you hope for?
- What is important for you when you think about the future? What would you like to do?
- Is there something you are worried about? What are you worried about?
- Is it something you can discuss with someone? If so, who?

Option 2: Patient says (s)he does not want to think about the future:

- What makes it difficult for you to think about the future?
- Do you know if XX (name FC) thinks about the future? What would (s)he thinks about? Does that keep you awake? Why (not)?

Thinking and talking about the future

Do you think about the care you might need in the future? What do think about? What are your expectations? What are your worries? What do you hope for?

- Who gave you this information?

Option 1: Patient indicates (s)he thinks about it:
- Would that be something you would like to discuss with someone? With whom?
- In case the patient does not wish to discuss future care with FC/HCP:
  - With whom you would like to discuss it? Why not with XX (name FC) or with HCP?
- In case the FC blocks the conversation about future/future care:
  - How do you feel about this?
  - What do you do when that happens?
  - What would you like to tell him/her?
- In case there is open communication about future/future care (see questions hereinafter).
- Patient indicates future or future care is not discussed, but believe the family carer might be open for it:
  - What do you hope to achieve with this conversation? What are your expectations?
  - What would you like to discuss?
  - How would that be for you to discuss your future/future care? Your worries, your ideas, your wishes, etc?
  - When would be the best timing to have this conversation?
  - Who should start the conversation? You, XX (name FC), HCP?
  - Would it be important to you to discuss this first privately with FC? Why (not)?
  - How do you think you would experience this conversation?

Option 2: Patient indicates (s)he does not wants to think about the future:
- What makes it difficult to think about the future?
- Does XX (name FC) has the same feeling?
- How would you react when XX (name FC) starts the conversation? Would you block it? Why (not)?

What are you afraid of that might happen if you talk about this?

Preferences about the future/future care

What is important for you when you think about your future or future care? Do you have specific wishes/ideas/preferences/expectations/hopes/etc + Ask more about the underlying values (What makes you have these wishes/ideas/XX; What is important for your when you think about your future or future care?).

You may give examples. ATTENTION: give only examples about what you have heard during the interview. How do you feel about a wheelchair, how do you feel about travelling, etc?

What would be the worst care for you and what would be the best care? Why?

Patient tell his wishes/preferences/ideas during the interview:

- Have you ever discussed these wishes, XX (give some examples about what they have said) with someone? Why (not)?

Option 1: patient indicates wishes/preferences were discussed during a conversation:

- Who was present during this conversation?
- What have you talked about?
- What has triggered the conversation?
    - Was the timing right? Why (not)?
    - In case the timing wasn't right: When would be the right timing for you?
- How did XX (name FC) think about your preferences/wishes?
- Did you talked about it several times after that conversation? Why (not)?
- Which decisions/plans were made about future/future care?
    - How did XX (name FC) felt about this?
    - Do you believe that soms preferences/wishes were difficult for XX (name FC)? Why (not)? What was difficult, what not? How do you cope with that?
- Did you ever consider writing these preferences/wishes down in a living will? Why (not)?
    - Do you believe that people close to you such as XX (name FC) knows which decisions they might need to make in case you cannot say them yourself? Why (not)?
- How did you end the conversation?
- What have you done afterwards?

Option 2: patient tells his/her wishes during interview, but indicates that (s)he did not discuss it with FC or HCP:

- Would you like to discuss your future/future care in the near or distant future? With whom?
- What is for you a barrier to have this conversation?
- What/who could help you to start this conversation? Would you like to discuss it with a HCP? If so, who?
- How would you feel, if you would want a conversation with XX (name FC) and you talk about XX (a preference that came forward during the interview), but XX (name FC) does not want it? OR How would you react if XX (name FC) would start a conversation about your future/future care?
- When would be the best timing to talk about the future or future care (for example to talk about preferred wheelchair)? Would this also be the right time to discuss XX (another preference)? Why (not)?

Option 3: patient makes it clear (s)he does not want to talk about future/future care/preferences/wishes/etc.

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

Option 1: In case future/future care has not been discussed yet with FC/HCP:

- When would be the right time for you?
- What makes it difficult to have this conversation?

Option 2: In case future/future care has been discussed:

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?

Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide family carers (FC): first interview

Introduction

XX (name patient) and you received XX months ago/ a while ago the diagnosis of ALS. Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first signals/symptoms?
- When did you notice that there was something wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you (as carer) the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

What do you know about the disease/trajectory of the disease? Which impact does ALS have?

- How did you receive this information? Who told you about this?
- What did you think/feel when you've heard this?

Do you think about your future and the future of XX (name patient)? Is it something that you worry about? Are you thinking more about the future than before the diagnosis, or vice versa, do you think about it less? Or is it another way of thinking about your future? Could you tell me something more about this?

Option 1: FC says (s)he thinks about the future:

- What do you expect? What do you hope for – for you and XX (name patient)?
- What is important for you when you think about the future? What do you think is important for the future of XX (name patient)?
- What makes you worry about the future?
- With whom would you prefer to discuss the future?

Option 2: FC thinks about the future, but they do not discuss it with the patient:

- Would you like to discuss it one day with XX (name patient)?
- Do you know what (s)he thinks about? Do you know what (s)he would want or hopes or expects about the future? What do you think is important for XX (name patient)?

Option 3: FC says they do not want to think about the future:

- You say you do not want to think about the future: How come? What makes you to refuse to think about the future?
- Is it something you are afraid of and why?

How do you think XX (name patient) thinks about it? Would (s)he thinks about the future? Would (s)he worries about it?

Thinking and talking about the future

Do you think about the care XX (name patient) might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for?

First asking the questions to see the perspective of the family carer AND then asking about how they think the patient thinks about these things.

- From whom did you receive information about his/her care?

Option 1: If FC thinks about future care:

- Is it something you discuss with XX (name patient)? Or with someone else? If so, who?
- If it is not possible to discuss care with patient/HCP:
    - Would you discuss it with friends/family/other professionals/etc?
- FC says (s)he tries to discuss it, but patient does not wish to have a conversation about his/her care:
    - How do you cope with it when you realize XX (name patient) does not want to talk about it?
    - What do you feel/think?
    - What would you like to say to XX (name patient)?
- FC indicates that they communicate openly about future care (go further to questions hereinafter).
- FC indicates they did not discuss future care, but thinks the patient might want to talk about it:
    - What do you hope this conversation would trigger? What are your expectations?
    - What would you like to say in that conversation?
    - How would that be to discuss your worries about XX (name patient) about his/her future?
    - When would it be important – according to you – to discuss it?
    - How would you experience such conversation?
    - Who should ideally start this conversation? Would you start it? Would you like that someone is there to support you? If so, who?
    - Would you prefer to have this discussion alone with XX (name patient) or together with a HCP? Why?

Option 2: FC indicates they do not wish to think about future or future care:

- Why do you not wish to think about future care?
- Does XX (name patient) share this opinion? How do you feel about the future care?
- How would you react/What would you do if XX (name patient) or a HCP wants to discuss future care and starts this conversation? Would you block it or would you talk about it? Why?

Preferences about the future/future care

What is important for you when you think about the future or future care of XX (name patient)? What do you wish for? What are your preferences/ideas? Are there things you know XX (name patient) would want or would not want about care? Are there things about care you do not want? What is – according to you important about XX's (name patient) future? What is important for you?

Here we can give examples they have indicated during the conversation or ask how they felt about the wheelchair or walking cane etc →* ***ATTENTION*: listen carefully what the FC says during the interview → you cannot talk about care wishes they did not talk about. So DON'T talk about a stomach-pump if they did not talk about it): For example you might ask: how do feel about the wheelchair? How do you think XX (name patient) feels about the wheelchair? OR I have heard you've made a lot of travels before your illness, is it something you wish to do with XX (name patient)? Do you think (s)he would enjoy this? Did you discuss it? Did you make plans? ***

OPTION 1: FC knows which preferences/wishes the patient has and they seem to talk about these preferences/wishes with one another:

- I hear that you have discuss it. Was there someone else present during this conversation? If so, who?
- What have you discussed?
- What did the conversation trigger? Did you believe the moment was ideal? Why?
    - In case they did not believe the timing was ideal: When would be the best timing?
- During the conversation, have your feelings been taken into account about certain wishes/preferences/ideas/expectations/hopes/etc XX (name patient) has? Did you say how you felt/thought about it? Why and how?
    - In case not: What makes you did not say how you felt? What might happen if you did?
- Did you make concrete plans about the future/future care?
- Have you considered to write these wishes/preferences/etc down? Why (not)?
    - How do you feel about a living will?
- Imagine you need to make a medical decision instead of XX (name patient), would you believe that you can make this decision? In other words, do you know what XX (name patient) wants or does not want?
- How did the conversation end? What have you done afterwards?

Option 2: FC seems to know about the preferences/wishes the patient have about the future/future care, but they did not discuss it concrete:

- What makes you so sure that XX (give example about a preferences/wish) is what XX (name patient) wants? Have you ever discussed it? Did you ever talk about before diagnosis? Or is it something that you see/feel? Would you like to elaborate this more?
- Would you like to have a conversation about the preferences/wishes with XX (name patient)? Why (not)?

  - What would you say or do if XX (name patient) says a wish/preference you Imagine that during this conversation XX (name patient) tells a wish/preference you cannot support?
  - When would be the ideal moment for you to have this discussion?
  - With whom you would like to discuss it? Only with XX (name patient) or also with HCP/kids/friends/etc?

OPTION 3: FC indicates (s)he does not know which preferences/wishes the patient has, but would like to know them:

- I hear you wish to have a conversation about the wishes/preferences XX (name patient) might have about the future/future care. Would this be something you would like to discuss alone with XX (name patient) or with a HCP or with kids/family/friends/etc?
- Would you initiate the conversation, or would you need help?
- Is there something that would make it difficult to discuss the wishes/preferences? If so, what? And what might help to overcome this barrier?
- What are the preferences you have about the future or future care of XX (name patient)?
- What do you hope to achieve in this conversation? What are your expectations?

OPTION 4: FC indicates (s)he does not know which preferences/wishes/etc the patient has, but FC does not want to know either:

- Why do you not want to know the preferences/wishes of XX (name patient)?

What is the worst possible thing that might happen if you would discuss it? Or if you would know the wishes/preferences of XX (name patient)?

Preferences about the conversation about future/future care

OPTION 1: FCs did not discuss wishes/preferences with patient/HCP/

- When would be the best timing to have this discussion with XX (name patient)?
- What makes it difficult to start this conversation?
- How do you think this conversation might go?
- How would you feel during such conversation?

Option 2: In case they have had this discussion:

- What triggered the conversation? How did it start?
- Who was present during this conversation? Was everyone present you and XX (name patient) would have wanted?
- Did someone help you with this conversation?
- How was the timing of this conversation? Did you feel the timing was right? Why (not)?
- How did you experience this conversation?
- Was everything discussed what you and XX (name patient) wished to discuss? Would you this conversation again in the future? Why (not)?

End of interview

Has everything been discussed what you’ve had liked to discuss?

Interview patient- FC together: first interview

Introduction

XX (name patient) and you received XX months ago/ a while ago the diagnosis of ALS. Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first signals/symptoms?
- When did you notice that there was something wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you (as patient and as carer) the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

What do you know about your disease/trajectory of the disease? Which impact does ALS have?

- How did you receive this information? Who told you about this?
- What did you think/feel when you've heard this?

Do you both think about your future? Is it something that you worry about? Are you thinking more about the future than before the diagnosis, or vice versa, do you think about it less? Or is it another way of thinking about your future? Could you tell me something more about this?

Option 1: Both telling that they think about the future:

- What do you expect? What do you hope for?
- What is important for you when you think about the future?
- What makes you worry about the future?
- With whom would you prefer to discuss the future?
- Do you discuss the future together? Why (not)?

Option 2: Both think about the future, but do not discuss this:

- Would you like to discuss it one day with each other? Why (not)?
- Do you know what (s)he thinks about? Do you know what (s)he would want or hopes or expects about the future? What do you think is important?

Option 3: In case one or both does not think about the future:

- You say you do not want to think about the future: How come? What makes you to refuse to think about the future?
- Is it something you are afraid of and why?

Thinking and talking about the future

Do you think about the care you might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for? <u>(patient)</u>

Do you, as a carer think about the care XX (name patient) might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for? <u>(family carer)</u>

- From whom did you receive information about the care?

Option 1: Both think about future care:
- Is it something you discuss together? Or with someone else? If so, who?
- Both indicate that they communicate openly about future care (go further to questions hereinafter).
- Both indicate they did not discuss future care, but both think about discussing it together:
    - ○ What do you hope this conversation would trigger? What are your expectations?
    - ○ What would you like to say in that conversation?
    - ○ How would that be to discuss your worries about XX (name patient) about his/her future and vice versa?
    - ○ When would it be important – according to you both – to discuss it?
    - ○ How would you experience such conversation?
    - ○ Who should ideally start this conversation? Would you start it? Would you like that someone is there to support you? If so, who?
    - ○ Would you prefer to have this discussion alone or together with a HCP? Why?

Option 2: Both indicate they do not wish to think about future or future care:
- Why do you not wish to think about future care?
- How do you feel about the future care?
- How would you react/What would you do if an HCP wants to discuss future care and starts this conversation? Would you block it or would you talk about it? Why?

Preferences about the future/future care

What is important for you when you think about the future or future care?
What do you wish for? What are your preferences/ideas? Are there things you would want or would not want about care? Are there things about care you do not want? <u>(patient)</u>

Here we can give examples they have indicated during the conversation or ask how they felt about the wheelchair or walking cane etc →* ***ATTENTION*: listen carefully what the FC says during the interview → you cannot talk about care wishes they did not talk about. So <u>DON'T</u> talk about a stomach-pump if they did not talk about it): For example you might ask: how do feel about the*

*wheelchair? OR I have heard you've made a lot of travels before your illness, is it something you wish to do? Did you discuss it? Did you make plans? ***

To family carer:

- What do you think, now you hear these preferences?
- How do you feel?
- Did you know that this was important for XX (name patient)? How did you know? Did you discuss this in the past? Why (not)?

OPTION 1: Patient expresses wishes and they have talked about these preferences/wishes with one another:

- I hear that you have discuss it. Was there someone else present during this conversation? If so, who?
- What have you discussed?
- What did the conversation trigger? Did you believe the moment was ideal? Why?
    - In case they did not believe the timing was ideal: When would be the best timing?
- During the conversation, have your feelings been taken into account about certain wishes/preferences/ideas/expectations/hopes/etc XX (name patient) has? Did you say how you felt/thought about it? Why and how?
    - In case not: What makes you did not say how you felt? What might happen if you did?
- Did you make concrete plans about the future/future care?
- Have you considered to write these wishes/preferences/etc down? Why (not)?
    - How do you feel about a living will?
- Imagine you need to make a medical decision instead of XX (name patient), would you believe that you can make this decision? In other words, do you know what XX (name patient) wants or does not want?
- How did the conversation end? What have you done afterwards?

OPTION 3: FC indicates (s)he did not know which preferences/wishes the patient has, but is glad that the patient gave some wishes and preferences during the interview:

- Do you believe that you would discuss these in more depth? Why (not)? Would this be something you would like to discuss alone with XX (name patient) or with a HCP or with kids/family/friends/etc?
- Who would initiate the conversation?
- What do you hope to achieve in this conversation? What are your expectations?

OPTION 4: Both did not say any wishes or preferences, and both indicate they do not wish to discuss it:

- Why do you not want to know the preferences/wishes of XX (name patient)?
- What is the worst possible thing that might happen if you would discuss it?

Preferences about the conversation about future/future care

OPTION 1: Both did not discuss wishes/preferences with patient/HCP/

- When would be the best timing to have this discussion with XX (name patient)?
- What makes it difficult to start this conversation?
- How do you think this conversation might go?
- How would you feel during such conversation?

Option 2: In case they have had this discussion:

- What triggered the conversation? How did it start?
- Who was present during this conversation? Was everyone present you and XX (name patient) would have wanted?
- Did someone help you with this conversation?
- How was the timing of this conversation? Did you feel the timing was right? Why (not)?
- How did you experience this conversation?
- Was everything discussed what you and XX (name patient) wished to discuss? Would you this conversation again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide for patient with ALS: subsequent interviews (interview 2 and 3)

NOTE: This interview guide only shows the main questions, because the subsequent interviews build upon what has been said in the previous interview.

Introduction

Could you tell me something more about the past 3 months, since our last conversation?

Did you think about our last conversation? Do you wish to clarify something?

What were for you the biggest changes since our last conversation? (*Changes physically, emotionally, socially)*

Thinking about the future

In the previous conversation, we have discussed thinking about the future. You told me XX (what they have told)

- How do you think about it now? Did it change? Why (not)?

Thinking and talking about the future

In the previous conversation, we have discussed talking about the future and then you have indicated you did (not) discuss the future with XX (name family carer) or with someone else

- Did you have this discussion in the past months? Why (not)?

OPTION 1: Patient did not talk about the future, and do not wish to talk about it:
- What makes it difficult to think about the future?
- Does XX (name FC) has the same feeling?
- How would you react when XX (name FC) starts the conversation? Would you block it? Why (not)?
- What are you afraid of that might happen if you talk about this?

OPTION 2: Patient thinks about the future, and wish to talk about the future, but family carer blocks the conversation:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?

OPTION 3: Patient thinks about the future, and has discussed it
- What triggered the conversation?

- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

- **NOTE:** ask how the previous interview influenced the possible conversation

Preferences about the future/future care

In the previous interview, we have discussed preferences about the future/future care. You said following things XX

- Did wishes/preferences/ideas change during these past months? Why (not)?
- In case wishes/preferences/ideas have changed
    - What triggered the change in your wishes/preferences/ideas?
    - Did you discuss these changes?
        - If so, with whom?
        - If not, why not?
- **NOTE:** ask how the previous interview influenced the possible change

In the previous interview, you told me that:

OPTION 1: you would want to discuss your preferences in the near future:

- Have you done this in the past months? Why (not)?
- In case family carers blocks the discussion:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?

OPTION 2: you have discussed your preferences prior to the first interview with XX (the persons they have indicated in the previous interview to whom they have discussed it)

- Have you discussed it again in the past months? Why (not)?

OPTION 3: I hear you have discussed your preferences in the past months

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

OPTION 4: you have not discussed your preferences and you do not wish to discuss this

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

In the previous conversation you have told me that the ideal timing for this conversation would be XX

OPTION 1: the ideal timing has arrived, but patient did not have this conversation

- Why did you not have this conversation? What stopped you?

OPTION 2: the ideal timing did not arrive, and there was no discussion in the meantime:

- Do you still have the same idea about the ideal timing to have this conversation? Why (not)?

OPTION 3: the ideal timing did not arrive, but patient did have a discussion in the past months:

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide for FC: subsequent interviews (interview 2 and 3)

NOTE: This interview guide only shows the main questions because the subsequent interviews build upon what has been said in the previous interview.

Introduction

Could you tell me something more about the past 3 months, since our last conversation?

Did you think about our last conversation? Do you wish to clarify something?

What were for you the biggest changes since our last conversation? (*Changes physically, emotionally, socially)*

Thinking about the future

In the previous conversation, we have discussed thinking about the future. You told me XX (what they have told)

- How do you think about it now? Did it change? Why (not)?

Thinking and talking about the future

In the previous conversation, we have discussed talking about the future and then you have indicated you did (not) discuss the future with XX (name patient) or with someone else

- Did you have this discussion in the past months? Why (not)?

OPTION 1: FC did not talk about the future, and do not wish to talk about it:

- What makes it difficult to think about the future?
- Does XX (name patient) has the same feeling?
- How would you react when XX (name patient) starts the conversation? Would you block it? Why (not)?
- What are you afraid of that might happen if you talk about this?

OPTION 2: FC thinks about the future, and wish to talk about the future, but patient blocks the conversation:

  - How do you feel about this?
  - What do you do when that happens?
  - What would you like to tell him/her?

OPTION 3: FC thinks about the future, and has discussed it

- What triggered the conversation?

- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

- **NOTE:** ask how the previous interview influenced the possible conversation

Preferences about the future/future care

In the previous interview, we have discussed preferences about the future/future care. You said following things XX

- Do you believe the wishes/preferences/ideas of XX (name patient) has changed during these past months? Why (not)?
- In case wishes/preferences/ideas have changed
    - What triggered the change in their wishes/preferences/ideas?
- **NOTE:** ask how the previous interview influenced the possible change

In the previous interview, you told me that:

OPTION 1: you would want to discuss the preferences with XX (name patient) in the near future:

- Have you done this in the past months? Why (not)?
- In case patient blocks the discussion:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?

OPTION 2: you have discussed with XX (name patient) the preferences prior to the first interview

- Have you discussed it again in the past months? Why (not)?

OPTION 3: I hear you have discussed the preferences with XX (name patient) in the past months

- What triggered the conversation?
- Who was present?
- Who initiated the conversation? You or XX (name patient) or someone else?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

OPTION 4: you have not discussed the preferences with XX (name patient) and you do not wish to discuss this

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

In the previous conversation you have told me that the ideal timing for this conversation would be XX

OPTION 1: the ideal timing has arrived, but FC did not have this conversation

- Why did you not have this conversation? What stopped you?

OPTION 2: the ideal timing did not arrive, and there was no discussion in the meantime:

- Do you still have the same idea about the ideal timing to have this conversation? Why (not)?

OPTION 3: the ideal timing did not arrive, but FC did have a discussion in the past months:

- What triggered the conversation?
- Who was present?
- Who initiated the conversation? You, XX (name patient) or someone else?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

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Brussel, 22 December 2021

Dear Editor,
Dear Prof dr Aldcroft,

Please find attached our study protocol titled “**Advance care planning in Amyotrophic Lateral Sclerosis (ALS): study protocol of a qualitative longitudinal study with persons with ALS and their family carers**”, which we submit for consideration for publication in BMJ open. This article describes the methodology on qualitative longitudinal multi-perspective interviews. In particular, we would like to inform international researchers on how to enhance the understanding in the experiences with ACP and the preferences for future (end-of-life) care and how these changes over time in a specific disease trajectory of ALS. Interviewing both perspectives of persons with ALS and their family carers will enlarge the understanding in dynamics and relationships between them, but also the individual needs in ACP for both persons with ALS and their family carers.

ALS is a degenerative motor neuron disease whereby the person with ALS endures progressive paralysis and dies within approximately 3 to 4 years after symptom onset, whereby death is most often caused by respiratory failure. There is no cure, no effective treatment and up to 50% of persons with ALS develop a cognitive impairment. This makes it clear that ACP cannot be delayed in this patient population. However, research of ACP in ALS is scarce and moreover, longitudinal qualitative multi-perspective studies in ACP are absent. Most studies investigate ACP from a single point in time, which only gives us a snapshot of the complex reality when engaging in ACP that is ideally done during multiple conversations throughout the disease trajectory. With this study we want to explore if, how and why ACP occurs and changes in a unique and fast-changing patient population of ALS and their family carers.

We will interview persons with ALS and their family carers individually to have both perspectives to explore similarities and differences in their views of ACP. This will result in high valuable information for clinical practice on how and when to implement ACP in care aligned with the individual needs, which will ultimately lead to better end-of-life care. We therefore believe that this work is of great interest to your international readership, and we hope that you will consider it for the review process in your journal.

For this study, ethical approval was obtained from the Ethical Review Board of Brussels University hospital of the Vrije Universiteit Brussel, and via amendment in the other participating hospitals (University Hospital Gent and AZ Maria Middelares) (B.U.N. B1432020000128)

We have no conflicts of interest to report. All listed authors contributed to the writing of the article and approved the final version of the manuscript.

If you require any further information, please do not hesitate to get in touch.

On behalf of all co-authors,
Yours sincerely,
Isabel Vandenbogaerde

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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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, multi-perspective design. A total of 8 to 9 dyads (pALS and their family carers) will be recruited, and semi-structured 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) website, social media and newsletter and via presentations at national and international scientific conferences.

Strengths and limitations of this study

- A longitudinal qualitative study design is an ideal method for capturing change in complex processes such as ACP
- Multi-perspective interviews will enhance our understanding of the dynamic relationships between pALS 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 to other longitudinal qualitative studies, but deemed feasible for addressing the aims of this exploratory study

Introduction

Amyotrophic lateral sclerosis (ALS) is the most common degenerative motor neuron disease (MND) in adults, affecting the brain and spinal cord.[1] ALS is incurable and characterized by progressive muscle paralysis. Respiratory failure is the most common cause of death.[2,3] The average survival between symptom onset and death is approximately three to four years, which is significantly shorter than the survival of people with other neurological conditions, such as dementia or multiple sclerosis.[1,4–7] 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.[8–11] 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[10–12]. 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[13].

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[14], especially if seen as a process with multiple conversations with patients and their family carers occurring over time[15]. However, the majority of studies have investigated ACP practice or participants' perceptions on ACP at one specific timepoint,[16–18] 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.[19]

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[8,9,20]. 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[21,22]. 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.[23] 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.[24,25] 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[26], 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)[27] and reduces the family carers' distress and grieving[14,28]. This shows that serial and multi-perspective 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 multi-perspective 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 3 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 people with ALS 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[29].

Methods

Study design

This study has a qualitative, longitudinal, multi-perspective interview design to provide rich information about the ACP process over time from the perspectives of both pALS and their family carers[30,31]. This design is most suitable for exploring an evolving and complex process such as ACP[32], as this method is driven by a desire to understand, not just if change happens, but how and why it happens in the socio-cultural context over time[32]. It offers considerable advantages over more typical 'snapshot' techniques in understanding the participants' changing experiences and preferences[31]. Constructionism will be used as an underlying epistemology,[33] 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 3 timepoints on a 3-monthly interval over a period of maximum 9 months. In Belgium, care for pALS is usually organized through neuromuscular reference centers, 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[34,35]. The neuromuscular reference centers are connected to University Hospitals. Care can also be provided by a general practitioner, community-based services (e.g., home care nurses) and palliative home care teams. Almost 90% of the patients stay at home even in the terminal stages of the disease[36]. Voluntary support services, in the form of national or regional ALS associations (e.g., ALS Liga in Belgium), can also deliver care to pALS and their families[3].

Participants and inclusion criteria

We will include patients from 3 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 multi-perspective study, we decided to include 8 to 9 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 3 times). This is a smaller sample compared to other longitudinal interview studies[37,38].

Recruitment

To recruit pALS and family carers, we have purposefully selected 2 academic hospitals (UZ Gent and UZ Brussel) and a non-academic hospital (AZ Maria Middelares). 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[39].

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 2 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 semi-structured interviews with persons with ALS and their family carers in Flanders, Belgium, at 3 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[30]. 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 upon 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 (e.g. unexpected hospitalization), and we will also reflect on whether and how the interviewer had an influence on the possible change (see Appendix I).

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[37,40]. 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 (e.g. 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[29]. 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[41,42]. Codes will be constructed in a coding list for each case separately for persons with ALS and family carers and from each interview round. In the case of any discrepancies, the codes will be discussed between 2 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.

Secondly, we will use a 2-step timeline method to describe changes in ACP experience and preferences within and over all the participants[43]. 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)[44]. To limit subjectivity, results of this timeline method will be discussed within the research team.

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 requested 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 (e.g., 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 emphasized 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.[29]

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.

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.

Discussion

This will be the first study to provide first-hand, longitudinal, in-depth, and multi-perspective 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[9,13] – 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[29,45]. 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 multi-perspective 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[37,40] and develop a trusting relationship with the interviewer,[29] which could limit participant attrition[40]. 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[46], 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[9,47] and they regard ACP as something beneficial[48,49]. 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 (8 to 9 dyads) compared to other longitudinal qualitative studies[37,38]. In Belgium, only 220 new ALS diagnoses occur per year[36], 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 8 to 9 dyads was deemed feasible for addressing the aims of this exploratory study. Hence, we will interpret our results with caution in terms of generalization to a larger group of pALS and their family carers.

Authors contribution

Conception and design of the work: IV, JDB, EC, AVDH, LVDB, LD, ADV; Ethic approval: IV, JDB, EC, AVDH, LVDB, LD, ADV; Drafting the work: IV; Critical revision for intellectual content: IV, RM, JDB, EC, AVDH, LVDB, LD, ADV; All authors have read and approved the final manuscript.

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Competing interests statement

The authors declare that they have no competing interests.

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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, multi-perspective design. A total of 8 to 9 dyads (pALS and their family carers) will be recruited, and ~~for~~ semi-structured interviews administered every 3 months over a~~for~~ 9 month period~~s~~. 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.

~~*Discussion:* This will be the first study to better understand the ACP communication process in ALS, which will inform clinical practice on how and when to implement ACP in the care of pALS and their families. The methodology described will inform other researchers on how to conduct longitudinal, multi-perspective qualitative interviews on ACP in other vulnerable patient populations.~~

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) website, social media and newsletter and via presentations at national and international scientific conferences.

Strengths and limitations of this study

- A longitudinal qualitative study design is an ideal method for capturing change in complex processes such as ACP
- Multi-perspective interviews will enhance our understanding of the dynamic relationships between pALS 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 to other longitudinal qualitative studies, but deemed feasible for addressing the aims of this exploratory study

Introduction

Amyotrophic lateral sclerosis (ALS) is the most common degenerative motor neuron disease (MND) in adults, affecting the brain and spinal cord.[1] ALS is incurable and characterized by progressive muscle paralysis. Respiratory failure is the most common cause of death.[2,3] The average survival between symptom onset and death is approximately three to four years, which is significantly shorter than the survival of people with other neurological conditions, such as dementia or multiple sclerosis.[1,4–7] 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.[8–11] 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[10–12]. 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[13].

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[14], especially if seen as a process with multiple conversations with patients and their family carers occurring over time[15]. However, the majority of studies have investigated ACP practice or participants' perceptions on ACP at one specific timepoint,[16–18] 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.[19]

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[8,9,20]. 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[21,22]. 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.[23] 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.[24,25] 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[26], 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)[27] and reduces the family carers' distress and grieving[14,28]. This shows that serial and multi-perspective 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 multi-perspective 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 3 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 people with ALS 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[29].

Methods

Study design

This study has a qualitative, longitudinal, multi-perspective interview design to provide rich information about the ACP process over time from the perspectives of both pALS and their family carers[30,31]. This design is most suitable for exploring an evolving and complex process such as ACP[32], as this method is driven by a desire to understand, not just if change happens, but how and why it happens in the socio-cultural context over time[32]. It offers considerable advantages over more typical 'snapshot' techniques in understanding the participants' changing experiences and preferences[31]. Constructionism will be used as an underlying epistemology,[33] 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 3 timepoints on a 3-monthly interval over a period of maximum 9 months. In Belgium, care for pALS is usually organized through neuromuscular reference centers, 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[34,35]. The neuromuscular reference centers are connected to University Hospitals. Care can also be provided by a general practitioner, community-based services (e.g., home care nurses) and palliative home care teams. Almost 90% of the patients stay at home even in the terminal stages of the disease[36]. Voluntary support services, in the form of national or regional ALS associations (e.g., ALS Liga in Belgium), can also deliver care to pALS and their families[3].

Participants and inclusion criteria

We will include patients from 3 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 multi-perspective study, we decided to include 8 to 9 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 3 times). This is a smaller sample compared to other longitudinal interview studies[37,38].

Recruitment

To recruit pALS and family carers, we have purposefully selected 2 academic hospitals (UZ Gent and UZ Brussel) and a non-academic hospital (AZ Maria Middelares). 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[39].

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 2 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 semi-structured interviews with persons with ALS and their family carers in Flanders, Belgium, at 3 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[30]. 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 upon 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 (e.g. unexpected hospitalization), and we will also reflect on whether and how the interviewer had an influence on the possible change (see Appendix I).

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[37,40]. 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 (e.g. 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[29]. In case of distress, the interviewer will advise the participants to talk to their ~~physician~~ 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[41,42]. Codes will be constructed in a coding list for each case separately for persons with ALS and family carers and from each interview round. In the case of any discrepancies, the codes will be discussed between 2 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.

Secondly, we will use a 2-step timeline method to describe changes in ACP experience and preferences within and over all the participants[43]. 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)[44]. To limit subjectivity, results of this timeline method will be discussed within the research team.

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 requested 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 (e.g., 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 emphasized 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.[29]

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.

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.

Discussion

This will be the first study to provide first-hand, longitudinal, in-depth, and multi-perspective 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[9,13] – 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[29,45]. 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 multi-perspective 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[37,40] and develop a trusting relationship with the interviewer,[29] which could limit participant attrition[40]. 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[46], 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[9,47] and they regard ACP as something beneficial[48,49]. 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 (8 to 9 dyads) compared to other longitudinal qualitative studies[37,38]. In Belgium, only 220 new ALS diagnoses occur per year[36], 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 8 to 9 dyads was deemed feasible for addressing the aims of this exploratory study. Hence, we will interpret our results with caution in terms of generalization to a larger group of pALS and their family carers.

~~Conclusion~~

~~This study will allow us to see in-depth if, how and why ACP occurs and changes in a unique patient population of ALS and their family carers. This protocol can inform other international researchers on how to conduct longitudinal, multi-perspective qualitative interviews in other populations who could also benefit from receiving ACP, but for whom the evidence concerning how ACP can be delivered optimally remains scarce.~~

Authors contribution

Conception and design of the work: IV, JDB, EC, AVDH, LVDB, LD, ADV; Ethic approval: IV, JDB, EC, AVDH, LVDB, LD, ADV; Drafting the work: IV; Critical revision for intellectual content: IV, RM, JDB, EC, AVDH, LVDB, LD, ADV; All authors have read and approved the final manuscript.

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The authors declare that they have no competing interests.

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Appendix I: interview guides

Interview guide 1: Person with ALS (interview 1)

Introduction

XX months ago you've received the diagnosis of ALS from XX (name neurologist). Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first symptoms/signals?
- When did you notice that something was wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

Can you tell me something more about ALS? What impact it has on you?

- Who told you this?
- What did you think/feel when you heard about this?

Do you think about your future? Is it something you think about it more than before the diagnosis or vice versa? How do you see your future now?

Option 1: Patient says (s)he thinks about the future:

- What are your expectations about the future? What do you hope for?
- What is important for you when you think about the future? What would you like to do?
- Is there something you are worried about? What are you worried about?
- Is it something you can discuss with someone? If so, who?

Option 2: Patient says (s)he does not want to think about the future:

- What makes it difficult for you to think about the future?
- Do you know if XX (name FC) thinks about the future? What would (s)he thinks about? Does that keep you awake? Why (not)?

Thinking and talking about the future

Do you think about the care you might need in the future? What do think about? What are your expectations? What are your worries? What do you hope for?

- Who gave you this information?

Option 1: Patient indicates (s)he thinks about it:

- Would that be something you would like to discuss with someone? With whom?
- In case the patient does not wish to discuss future care with FC/HCP:
    - With whom you would like to discuss it? Why not with XX (name FC) or with HCP?
- In case the FC blocks the conversation about future/future care:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?
- In case there is open communication about future/future care (see questions hereinafter).
- Patient indicates future or future care is not discussed, but believe the family carer might be open for it:
    - What do you hope to achieve with this conversation? What are your expectations?
    - What would you like to discuss?
    - How would that be for you to discuss your future/future care? Your worries, your ideas, your wishes, etc?
    - When would be the best timing to have this conversation?
    - Who should start the conversation? You, XX (name FC), HCP?
    - Would it be important to you to discuss this first privately with FC? Why (not)?
    - How do you think you would experience this conversation?

Option 2: Patient indicates (s)he does not wants to think about the future:

- What makes it difficult to think about the future?
- Does XX (name FC) has the same feeling?
- How would you react when XX (name FC) starts the conversation? Would you block it? Why (not)?

What are you afraid of that might happen if you talk about this?

Preferences about the future/future care

What is important for you when you think about your future or future care? Do you have specific wishes/ideas/preferences/expectations/hopes/etc + Ask more about the underlying values (What makes you have these wishes/ideas/XX; What is important for your when you think about your future or future care?).

You may give examples. ATTENTION: give only examples about what you have heard during the interview. How do you feel about a wheelchair, how do you feel about travelling, etc?

What would be the worst care for you and what would be the best care? Why?

Patient tell his wishes/preferences/ideas during the interview:

- Have you ever discussed these wishes, XX (give some examples about what they have said) with someone? Why (not)?

Option 1: patient indicates wishes/preferences were discussed during a conversation:

- Who was present during this conversation?
- What have you talked about?
- What has triggered the conversation?
    - Was the timing right? Why (not)?
    - In case the timing wasn't right: When would be the right timing for you?
- How did XX (name FC) think about your preferences/wishes?
- Did you talked about it several times after that conversation? Why (not)?
- Which decisions/plans were made about future/future care?
    - How did XX (name FC) felt about this?
    - Do you believe that soms preferences/wishes were difficult for XX (name FC)? Why (not)? What was difficult, what not? How do you cope with that?
- Did you ever consider writing these preferences/wishes down in a living will? Why (not)?
    - Do you believe that people close to you such as XX (name FC) knows which decisions they might need to make in case you cannot say them yourself? Why (not)?
- How did you end the conversation?
- What have you done afterwards?

Option 2: patient tells his/her wishes during interview, but indicates that (s)he did not discuss it with FC or HCP:

- Would you like to discuss your future/future care in the near or distant future? With whom?
- What is for you a barrier to have this conversation?
- What/who could help you to start this conversation? Would you like to discuss it with a HCP? If so, who?
- How would you feel, if you would want a conversation with XX (name FC) and you talk about XX (a preference that came forward during the interview), but XX (name FC) does not want it? OR How would you react if XX (name FC) would start a conversation about your future/future care?
- When would be the best timing to talk about the future or future care (for example to talk about preferred wheelchair)? Would this also be the right time to discuss XX (another preference)? Why (not)?

Option 3: patient makes it clear (s)he does not want to talk about future/future care/preferences/wishes/etc.

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

Option 1: In case future/future care has not been discussed yet with FC/HCP:

- When would be the right time for you?
- What makes it difficult to have this conversation?

Option 2: In case future/future care has been discussed:

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?

Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide family carers (FC): first interview

Introduction

XX (name patient) and you received XX months ago/ a while ago the diagnosis of ALS. Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first signals/symptoms?
- When did you notice that there was something wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you (as carer) the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

What do you know about the disease/trajectory of the disease? Which impact does ALS have?

- How did you receive this information? Who told you about this?
- What did you think/feel when you've heard this?

Do you think about your future and the future of XX (name patient)? Is it something that you worry about? Are you thinking more about the future than before the diagnosis, or vice versa, do you think about it less? Or is it another way of thinking about your future? Could you tell me something more about this?

Option 1: FC says (s)he thinks about the future:
- What do you expect? What do you hope for – for you and XX (name patient)?
- What is important for you when you think about the future? What do you think is important for the future of XX (name patient)?
- What makes you worry about the future?
- With whom would you prefer to discuss the future?

Option 2: FC thinks about the future, but they do not discuss it with the patient:
- Would you like to discuss it one day with XX (name patient)?
- Do you know what (s)he thinks about? Do you know what (s)he would want or hopes or expects about the future? What do you think is important for XX (name patient)?

Option 3: FC says they do not want to think about the future:
- You say you do not want to think about the future: How come? What makes you to refuse to think about the future?
- Is it something you are afraid of and why?

How do you think XX (name patient) thinks about it? Would (s)he thinks about the future? Would (s)he worries about it?

Thinking and talking about the future

Do you think about the care XX (name patient) might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for?

First asking the questions to see the perspective of the family carer AND then asking about how they think the patient thinks about these things.

- From whom did you receive information about his/her care?

Option 1: If FC thinks about future care:

- Is it something you discuss with XX (name patient)? Or with someone else? If so, who?
- If it is not possible to discuss care with patient/HCP:
    - Would you discuss it with friends/family/other professionals/etc?
- FC says (s)he tries to discuss it, but patient does not wish to have a conversation about his/her care:
    - How do you cope with it when you realize XX (name patient) does not want to talk about it?
    - What do you feel/think?
    - What would you like to say to XX (name patient)?
- FC indicates that they communicate openly about future care (go further to questions hereinafter).
- FC indicates they did not discuss future care, but thinks the patient might want to talk about it:
    - What do you hope this conversation would trigger? What are your expectations?
    - What would you like to say in that conversation?
    - How would that be to discuss your worries about XX (name patient) about his/her future?
    - When would it be important – according to you – to discuss it?
    - How would you experience such conversation?
    - Who should ideally start this conversation? Would you start it? Would you like that someone is there to support you? If so, who?
    - Would you prefer to have this discussion alone with XX (name patient) or together with a HCP? Why?

Option 2: FC indicates they do not wish to think about future or future care:

- Why do you not wish to think about future care?
- Does XX (name patient) share this opinion? How do you feel about the future care?
- How would you react/What would you do if XX (name patient) or a HCP wants to discuss future care and starts this conversation? Would you block it or would you talk about it? Why?

Preferences about the future/future care

What is important for you when you think about the future or future care of XX (name patient)? What do you wish for? What are your preferences/ideas? Are there things you know XX (name patient) would want or would not want about care? Are there things about care you do not want? What is – according to you important about XX's (name patient) future? What is important for you?

***Here we can give examples they have indicated during the conversation or ask how they felt about the wheelchair or walking cane etc → **ATTENTION**: listen carefully what the FC says during the interview → you cannot talk about care wishes they did not talk about. So <u>DON'T</u> talk about a stomach-pump if they did not talk about it): For example you might ask: how do feel about the wheelchair? How do you think XX (name patient) feels about the wheelchair? OR I have heard you've made a lot of travels before your illness, is it something you wish to do with XX (name patient)? Do you think (s)he would enjoy this? Did you discuss it? Did you make plans? ***

OPTION 1: FC knows which preferences/wishes the patient has and they seem to talk about these preferences/wishes with one another:

- I hear that you have discuss it. Was there someone else present during this conversation? If so, who?
- What have you discussed?
- What did the conversation trigger? Did you believe the moment was ideal? Why?
    - In case they did not believe the timing was ideal: When would be the best timing?
- During the conversation, have your feelings been taken into account about certain wishes/preferences/ideas/expectations/hopes/etc XX (name patient) has? Did you say how you felt/thought about it? Why and how?
    - In case not: What makes you did not say how you felt? What might happen if you did?
- Did you make concrete plans about the future/future care?
- Have you considered to write these wishes/preferences/etc down? Why (not)?
    - How do you feel about a living will?
- Imagine you need to make a medical decision instead of XX (name patient), would you believe that you can make this decision? In other words, do you know what XX (name patient) wants or does not want?
- How did the conversation end? What have you done afterwards?

Option 2: FC seems to know about the preferences/wishes the patient have about the future/future care, but they did not discuss it concrete:

- What makes you so sure that XX (give example about a preferences/wish) is what XX (name patient) wants? Have you ever discussed it? Did you ever talk about before diagnosis? Or is it something that you see/feel? Would you like to elaborate this more?
- Would you like to have a conversation about the preferences/wishes with XX (name patient)? Why (not)?

    - What would you say or do if XX (name patient) says a wish/preference you Imagine that during this conversation XX (name patient) tells a wish/preference you cannot support?
    - When would be the ideal moment for you to have this discussion?
    - With whom you would like to discuss it? Only with XX (name patient) or also with HCP/kids/friends/etc?

OPTION 3: FC indicates (s)he does not know which preferences/wishes the patient has, but would like to know them:

- I hear you wish to have a conversation about the wishes/preferences XX (name patient) might have about the future/future care. Would this be something you would like to discuss alone with XX (name patient) or with a HCP or with kids/family/friends/etc?
- Would you initiate the conversation, or would you need help?
- Is there something that would make it difficult to discuss the wishes/preferences? If so, what? And what might help to overcome this barrier?
- What are the preferences you have about the future or future care of XX (name patient)?
- What do you hope to achieve in this conversation? What are your expectations?

OPTION 4: FC indicates (s)he does not know which preferences/wishes/etc the patient has, but FC does not want to know either:

- Why do you not want to know the preferences/wishes of XX (name patient)?

What is the worst possible thing that might happen if you would discuss it? Or if you would know the wishes/preferences of XX (name patient)?

Preferences about the conversation about future/future care

OPTION 1: FCs did not discuss wishes/preferences with patient/HCP/

- When would be the best timing to have this discussion with XX (name patient)?
- What makes it difficult to start this conversation?
- How do you think this conversation might go?
- How would you feel during such conversation?

Option 2: In case they have had this discussion:

- What triggered the conversation? How did it start?
- Who was present during this conversation? Was everyone present you and XX (name patient) would have wanted?
- Did someone help you with this conversation?
- How was the timing of this conversation? Did you feel the timing was right? Why (not)?
- How did you experience this conversation?
- Was everything discussed what you and XX (name patient) wished to discuss? Would you this conversation again in the future? Why (not)?

End of interview

Has everything been discussed what you’ve had liked to discuss?

Interview patient- FC together: first interview

Introduction

XX (name patient) and you received XX months ago/ a while ago the diagnosis of ALS. Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first signals/symptoms?
- When did you notice that there was something wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you (as patient and as carer) the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

What do you know about your disease/trajectory of the disease? Which impact does ALS have?

- How did you receive this information? Who told you about this?
- What did you think/feel when you've heard this?

Do you both think about your future? Is it something that you worry about? Are you thinking more about the future than before the diagnosis, or vice versa, do you think about it less? Or is it another way of thinking about your future? Could you tell me something more about this?

Option 1: Both telling that they think about the future:

- What do you expect? What do you hope for?
- What is important for you when you think about the future?
- What makes you worry about the future?
- With whom would you prefer to discuss the future?
- Do you discuss the future together? Why (not)?

Option 2: Both think about the future, but do not discuss this:

- Would you like to discuss it one day with each other? Why (not)?
- Do you know what (s)he thinks about? Do you know what (s)he would want or hopes or expects about the future? What do you think is important?

Option 3: In case one or both does not think about the future:

- You say you do not want to think about the future: How come? What makes you to refuse to think about the future?
- Is it something you are afraid of and why?

Thinking and talking about the future

Do you think about the care you might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for? <u>(patient)</u>

Do you, as a carer think about the care XX (name patient) might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for? <u>(family carer)</u>

- From whom did you receive information about the care?

Option 1: Both think about future care:

- Is it something you discuss together? Or with someone else? If so, who?
- Both indicate that they communicate openly about future care (go further to questions hereinafter).
- Both indicate they did not discuss future care, but both think about discussing it together:
    - What do you hope this conversation would trigger? What are your expectations?
    - What would you like to say in that conversation?
    - How would that be to discuss your worries about XX (name patient) about his/her future and vice versa?
    - When would it be important – according to you both – to discuss it?
    - How would you experience such conversation?
    - Who should ideally start this conversation? Would you start it? Would you like that someone is there to support you? If so, who?
    - Would you prefer to have this discussion alone or together with a HCP? Why?

Option 2: Both indicate they do not wish to think about future or future care:

- Why do you not wish to think about future care?
- How do you feel about the future care?
- How would you react/What would you do if an HCP wants to discuss future care and starts this conversation? Would you block it or would you talk about it? Why?

Preferences about the future/future care

What is important for you when you think about the future or future care?
What do you wish for? What are your preferences/ideas? Are there things you would want or would not want about care? Are there things about care you do not want? <u>(patient)</u>

***Here we can give examples they have indicated during the conversation or ask how they felt about the wheelchair or walking cane etc → **ATTENTION**: listen carefully what the FC says during the interview → you cannot talk about care wishes they did not talk about. So <u>DON'T</u> talk about a stomach-pump if they did not talk about it): For example you might ask: how do feel about the*

*wheelchair? OR I have heard you've made a lot of travels before your illness, is it something you wish to do? Did you discuss it? Did you make plans? ***

To family carer:

- What do you think, now you hear these preferences?
- How do you feel?
- Did you know that this was important for XX (name patient)? How did you know? Did you discuss this in the past? Why (not)?

OPTION 1: Patient expresses wishes and they have talked about these preferences/wishes with one another:

- I hear that you have discuss it. Was there someone else present during this conversation? If so, who?
- What have you discussed?
- What did the conversation trigger? Did you believe the moment was ideal? Why?
    - In case they did not believe the timing was ideal: When would be the best timing?
- During the conversation, have your feelings been taken into account about certain wishes/preferences/ideas/expectations/hopes/etc XX (name patient) has? Did you say how you felt/thought about it? Why and how?
    - In case not: What makes you did not say how you felt? What might happen if you did?
- Did you make concrete plans about the future/future care?
- Have you considered to write these wishes/preferences/etc down? Why (not)?
    - How do you feel about a living will?
- Imagine you need to make a medical decision instead of XX (name patient), would you believe that you can make this decision? In other words, do you know what XX (name patient) wants or does not want?
- How did the conversation end? What have you done afterwards?

OPTION 3: FC indicates (s)he did not know which preferences/wishes the patient has, but is glad that the patient gave some wishes and preferences during the interview:

- Do you believe that you would discuss these in more depth? Why (not)? Would this be something you would like to discuss alone with XX (name patient) or with a HCP or with kids/family/friends/etc?
- Who would initiate the conversation?
- What do you hope to achieve in this conversation? What are your expectations?

OPTION 4: Both did not say any wishes or preferences, and both indicate they do not wish to discuss it:

- Why do you not want to know the preferences/wishes of XX (name patient)?
- What is the worst possible thing that might happen if you would discuss it?

Preferences about the conversation about future/future care

OPTION 1: Both did not discuss wishes/preferences with patient/HCP/

- When would be the best timing to have this discussion with XX (name patient)?
- What makes it difficult to start this conversation?
- How do you think this conversation might go?
- How would you feel during such conversation?

Option 2: In case they have had this discussion:

- What triggered the conversation? How did it start?
- Who was present during this conversation? Was everyone present you and XX (name patient) would have wanted?
- Did someone help you with this conversation?
- How was the timing of this conversation? Did you feel the timing was right? Why (not)?
- How did you experience this conversation?
- Was everything discussed what you and XX (name patient) wished to discuss? Would you this conversation again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide for patient with ALS: subsequent interviews (interview 2 and 3)

NOTE: This interview guide only shows the main questions, because the subsequent interviews build upon what has been said in the previous interview.

Introduction

Could you tell me something more about the past 3 months, since our last conversation?

Did you think about our last conversation? Do you wish to clarify something?

What were for you the biggest changes since our last conversation? (*Changes physically, emotionally, socially)*

Thinking about the future

In the previous conversation, we have discussed thinking about the future. You told me XX (what they have told)

- How do you think about it now? Did it change? Why (not)?

Thinking and talking about the future

In the previous conversation, we have discussed talking about the future and then you have indicated you did (not) discuss the future with XX (name family carer) or with someone else

- Did you have this discussion in the past months? Why (not)?

OPTION 1: Patient did not talk about the future, and do not wish to talk about it:

- What makes it difficult to think about the future?
- Does XX (name FC) has the same feeling?
- How would you react when XX (name FC) starts the conversation? Would you block it? Why (not)?
- What are you afraid of that might happen if you talk about this?

OPTION 2: Patient thinks about the future, and wish to talk about the future, but family carer blocks the conversation:

    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?

OPTION 3: Patient thinks about the future, and has discussed it

- What triggered the conversation?

- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

- **NOTE:** ask how the previous interview influenced the possible conversation

Preferences about the future/future care

In the previous interview, we have discussed preferences about the future/future care. You said following things XX

- Did wishes/preferences/ideas change during these past months? Why (not)?
- In case wishes/preferences/ideas have changed
    - What triggered the change in your wishes/preferences/ideas?
    - Did you discuss these changes?
        - If so, with whom?
        - If not, why not?
- **NOTE:** ask how the previous interview influenced the possible change

In the previous interview, you told me that:

OPTION 1: you would want to discuss your preferences in the near future:

- Have you done this in the past months? Why (not)?
- In case family carers blocks the discussion:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?

OPTION 2: you have discussed your preferences prior to the first interview with XX (the persons they have indicated in the previous interview to whom they have discussed it)

- Have you discussed it again in the past months? Why (not)?

OPTION 3: I hear you have discussed your preferences in the past months

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

OPTION 4: you have not discussed your preferences and you do not wish to discuss this

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

In the previous conversation you have told me that the ideal timing for this conversation would be XX

OPTION 1: the ideal timing has arrived, but patient did not have this conversation

- Why did you not have this conversation? What stopped you?

OPTION 2: the ideal timing did not arrive, and there was no discussion in the meantime:

- Do you still have the same idea about the ideal timing to have this conversation? Why (not)?

OPTION 3: the ideal timing did not arrive, but patient did have a discussion in the past months:

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you’ve had liked to discuss?

Interview guide for FC: subsequent interviews (interview 2 and 3)

NOTE: This interview guide only shows the main questions because the subsequent interviews build upon what has been said in the previous interview.

Introduction

Could you tell me something more about the past 3 months, since our last conversation?

Did you think about our last conversation? Do you wish to clarify something?

What were for you the biggest changes since our last conversation? (*Changes physically, emotionally, socially)*

Thinking about the future

In the previous conversation, we have discussed thinking about the future. You told me XX (what they have told)

- How do you think about it now? Did it change? Why (not)?

Thinking and talking about the future

In the previous conversation, we have discussed talking about the future and then you have indicated you did (not) discuss the future with XX (name patient) or with someone else

- Did you have this discussion in the past months? Why (not)?

OPTION 1: FC did not talk about the future, and do not wish to talk about it:

- What makes it difficult to think about the future?
- Does XX (name patient) has the same feeling?
- How would you react when XX (name patient) starts the conversation? Would you block it? Why (not)?
- What are you afraid of that might happen if you talk about this?

OPTION 2: FC thinks about the future, and wish to talk about the future, but patient blocks the conversation:

  - How do you feel about this?
  - What do you do when that happens?
  - What would you like to tell him/her?

OPTION 3: FC thinks about the future, and has discussed it

- What triggered the conversation?

- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

- **NOTE:** ask how the previous interview influenced the possible conversation

Preferences about the future/future care

In the previous interview, we have discussed preferences about the future/future care. You said following things XX

- Do you believe the wishes/preferences/ideas of XX (name patient) has changed during these past months? Why (not)?
- In case wishes/preferences/ideas have changed
  - What triggered the change in their wishes/preferences/ideas?
- **NOTE:** ask how the previous interview influenced the possible change

In the previous interview, you told me that:

OPTION 1: you would want to discuss the preferences with XX (name patient) in the near future:

- Have you done this in the past months? Why (not)?
- In case patient blocks the discussion:
  - How do you feel about this?
  - What do you do when that happens?
  - What would you like to tell him/her?

OPTION 2: you have discussed with XX (name patient) the preferences prior to the first interview

- Have you discussed it again in the past months? Why (not)?

OPTION 3: I hear you have discussed the preferences with XX (name patient) in the past months

- What triggered the conversation?
- Who was present?
- Who initiated the conversation? You or XX (name patient) or someone else?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

OPTION 4: you have not discussed the preferences with XX (name patient) and you do not wish to discuss this

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

In the previous conversation you have told me that the ideal timing for this conversation would be XX

OPTION 1: the ideal timing has arrived, but FC did not have this conversation

- Why did you not have this conversation? What stopped you?

OPTION 2: the ideal timing did not arrive, and there was no discussion in the meantime:

- Do you still have the same idea about the ideal timing to have this conversation? Why (not)?

OPTION 3: the ideal timing did not arrive, but FC did have a discussion in the past months:

- What triggered the conversation?
- Who was present?
- Who initiated the conversation? You, XX (name patient) or someone else?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Note from the Editors: Instructions for reviewers of study protocols

Since launching in 2011, BMJ Open has published study protocols for planned or ongoing research studies. If data collection is complete, we will not consider the manuscript.

Publishing study protocols enables researchers and funding bodies to stay up to date in their fields by providing exposure to research activity that may not otherwise be widely publicised. This can help prevent unnecessary duplication of work and will hopefully enable collaboration. Publishing protocols in full also makes available more information than is currently required by trial registries and increases transparency, making it easier for others (editors, reviewers and readers) to see and understand any deviations from the protocol that occur during the conduct of the study.

The scientific integrity and the credibility of the study data depend substantially on the study design and methodology, which is why the study protocol requires a thorough peer-review.

BMJ Open will consider for publication protocols for any study design, including observational studies and systematic reviews.

Some things to keep in mind when reviewing the study protocol:

- Protocol papers should report planned or ongoing studies. The dates of the study should be included in the manuscript.
- Unfortunately we are unable to customize the reviewer report form for study protocols. As such, some of the items (i.e., those pertaining to results) on the form should be scores as Not Applicable (N/A).
- While some baseline data can be presented, there should be no results or conclusions present in the study protocol.
- For studies that are ongoing, it is generally the case that very few changes can be made to the methodology. As such, requests for revisions are generally clarifications for the rationale or details relating to the methods. If there is a major flaw in the study that would prevent a sound interpretation of the data, we would expect the study protocol to be rejected.

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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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, multi-perspective design. A total of 8 to 9 dyads (pALS and their family carers) will be recruited, and semi-structured 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) website, social media and newsletter and via presentations at national and international scientific conferences.

Strengths and limitations of this study

- A longitudinal qualitative study design is an ideal method for capturing change in complex processes such as ACP
- Multi-perspective interviews will enhance our understanding of the dynamic relationships between pALS 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 to other longitudinal qualitative studies, but deemed feasible for addressing the aims of this exploratory study

Introduction

Amyotrophic lateral sclerosis (ALS) is the most common degenerative motor neuron disease (MND) in adults, affecting the brain and spinal cord.[1] ALS is incurable and characterized by progressive muscle paralysis. Respiratory failure is the most common cause of death.[2,3] The average survival between symptom onset and death is approximately three to four years, which is significantly shorter than the survival of people with other neurological conditions, such as dementia or multiple sclerosis.[1,4–7] 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.[8–11] 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[10–12]. 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[13].

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[14], especially if seen as a process with multiple conversations with patients and their family carers occurring over time[15]. However, the majority of studies have investigated ACP practice or participants' perceptions on ACP at one specific timepoint,[16–18] 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.[19]

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[8,9,20]. 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[21,22]. 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.[23] 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.[24,25] 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[26], 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)[27] and reduces the family carers' distress and grieving[14,28]. This shows that serial and multi-perspective 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 multi-perspective 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 3 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 people with ALS 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[29].

Methods

Study design

This study has a qualitative, longitudinal, multi-perspective interview design to provide rich information about the ACP process over time from the perspectives of both pALS and their family carers[30,31]. This design is most suitable for exploring an evolving and complex process such as ACP[32], as this method is driven by a desire to understand, not just if change happens, but how and why it happens in the socio-cultural context over time[32]. It offers considerable advantages over more typical 'snapshot' techniques in understanding the participants' changing experiences and preferences[31]. Constructionism will be used as an underlying epistemology,[33] 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 3 timepoints on a 3-monthly interval over a period of maximum 9 months. In Belgium, care for pALS is usually organized through neuromuscular reference centers, 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[34,35]. The neuromuscular reference centers are connected to University Hospitals. Care can also be provided by a general practitioner, community-based services (e.g., home care nurses) and palliative home care teams. Almost 90% of the patients stay at home even in the terminal stages of the disease[36]. Voluntary support services, in the form of national or regional ALS associations (e.g., ALS Liga in Belgium), can also deliver care to pALS and their families[3].

Participants and inclusion criteria

We will include patients from 3 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 multi-perspective study, we decided to include 8 to 9 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 3 times). This is a smaller sample compared to other longitudinal interview studies[37,38].

Recruitment

To recruit pALS and family carers, we have purposefully selected 2 academic hospitals (UZ Gent and UZ Brussel) and a non-academic hospital (AZ Maria Middelares). 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[39].

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 2 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 semi-structured interviews with persons with ALS and their family carers in Flanders, Belgium, at 3 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[30]. 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 upon 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 (e.g. unexpected hospitalization), and we will also reflect on whether and how the interviewer had an influence on the possible change (see Appendix I).

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[37,40]. 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 (e.g. 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[29]. 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[41,42]. Codes will be constructed in a coding list for each case separately for persons with ALS and family carers and from each interview round. In the case of any discrepancies, the codes will be discussed between 2 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.

Secondly, we will use a 2-step timeline method to describe changes in ACP experience and preferences within and over all the participants[43]. 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)[44]. 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 (e.g., 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 emphasized 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.[29]

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 multi-perspective 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[9,13] – 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[29,45]. 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 multi-perspective 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[37,40] and develop a trusting relationship with the interviewer,[29] which could limit participant attrition[40]. 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[46], 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[9,47] and they regard ACP as something beneficial[48,49]. 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 (8 to 9 dyads) compared to other longitudinal qualitative studies[37,38]. In Belgium, only 220 new ALS diagnoses occur per year[36], 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 8 to 9 dyads was deemed feasible for addressing the aims of this exploratory study. Hence, we will interpret our results with caution in terms of generalization to a larger group of pALS and their family carers.

Authors contribution

Conception and design of the work: IV, JDB, EC, AVDH, LVDB, LD, ADV; Ethic approval: IV, JDB, EC, AVDH, LVDB, LD, ADV; Drafting the work: IV; Critical revision for intellectual content: IV, RM, JDB, EC, AVDH, LVDB, LD, ADV; All authors have read and approved the final manuscript.

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Competing interests statement

The authors declare that they have no competing interests.

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Appendix I: interview guides

Interview guide 1: Person with ALS (interview 1)

Introduction

XX months ago you've received the diagnosis of ALS from XX (name neurologist). Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first symptoms/signals?
- When did you notice that something was wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

Can you tell me something more about ALS? What impact it has on you?

- Who told you this?
- What did you think/feel when you heard about this?

Do you think about your future? Is it something you think about it more than before the diagnosis or vice versa? How do you see your future now?

Option 1: Patient says (s)he thinks about the future:

- What are your expectations about the future? What do you hope for?
- What is important for you when you think about the future? What would you like to do?
- Is there something you are worried about? What are you worried about?
- Is it something you can discuss with someone? If so, who?

Option 2: Patient says (s)he does not want to think about the future:

- What makes it difficult for you to think about the future?
- Do you know if XX (name FC) thinks about the future? What would (s)he thinks about? Does that keep you awake? Why (not)?

Thinking and talking about the future

Do you think about the care you might need in the future? What do think about? What are your expectations? What are your worries? What do you hope for?

- Who gave you this information?

Option 1: Patient indicates (s)he thinks about it:

- Would that be something you would like to discuss with someone? With whom?
- In case the patient does not wish to discuss future care with FC/HCP:
    - With whom you would like to discuss it? Why not with XX (name FC) or with HCP?
- In case the FC blocks the conversation about future/future care:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?
- In case there is open communication about future/future care (see questions hereinafter).
- Patient indicates future or future care is not discussed, but believe the family carer might be open for it:
    - What do you hope to achieve with this conversation? What are your expectations?
    - What would you like to discuss?
    - How would that be for you to discuss your future/future care? Your worries, your ideas, your wishes, etc?
    - When would be the best timing to have this conversation?
    - Who should start the conversation? You, XX (name FC), HCP?
    - Would it be important to you to discuss this first privately with FC? Why (not)?
    - How do you think you would experience this conversation?

Option 2: Patient indicates (s)he does not wants to think about the future:

- What makes it difficult to think about the future?
- Does XX (name FC) has the same feeling?
- How would you react when XX (name FC) starts the conversation? Would you block it? Why (not)?

What are you afraid of that might happen if you talk about this?

Preferences about the future/future care

What is important for you when you think about your future or future care? Do you have specific wishes/ideas/preferences/expectations/hopes/etc + Ask more about the underlying values (What makes you have these wishes/ideas/XX; What is important for your when you think about your future or future care?).

You may give examples. ATTENTION: give only examples about what you have heard during the interview. How do you feel about a wheelchair, how do you feel about travelling, etc?

What would be the worst care for you and what would be the best care? Why?

Patient tell his wishes/preferences/ideas during the interview:

- Have you ever discussed these wishes, XX (give some examples about what they have said) with someone? Why (not)?

Option 1: patient indicates wishes/preferences were discussed during a conversation:

- Who was present during this conversation?
- What have you talked about?
- What has triggered the conversation?
    - Was the timing right? Why (not)?
    - In case the timing wasn't right: When would be the right timing for you?
- How did XX (name FC) think about your preferences/wishes?
- Did you talked about it several times after that conversation? Why (not)?
- Which decisions/plans were made about future/future care?
    - How did XX (name FC) felt about this?
    - Do you believe that soms preferences/wishes were difficult for XX (name FC)? Why (not)? What was difficult, what not? How do you cope with that?
- Did you ever consider writing these preferences/wishes down in a living will? Why (not)?
    - Do you believe that people close to you such as XX (name FC) knows which decisions they might need to make in case you cannot say them yourself? Why (not)?
- How did you end the conversation?
- What have you done afterwards?

Option 2: patient tells his/her wishes during interview, but indicates that (s)he did not discuss it with FC or HCP:

- Would you like to discuss your future/future care in the near or distant future? With whom?
- What is for you a barrier to have this conversation?
- What/who could help you to start this conversation? Would you like to discuss it with a HCP? If so, who?
- How would you feel, if you would want a conversation with XX (name FC) and you talk about XX (a preference that came forward during the interview), but XX (name FC) does not want it? OR How would you react if XX (name FC) would start a conversation about your future/future care?
- When would be the best timing to talk about the future or future care (for example to talk about preferred wheelchair)? Would this also be the right time to discuss XX (another preference)? Why (not)?

Option 3: patient makes it clear (s)he does not want to talk about future/future care/preferences/wishes/etc.

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

Option 1: In case future/future care has not been discussed yet with FC/HCP:

- When would be the right time for you?
- What makes it difficult to have this conversation?

Option 2: In case future/future care has been discussed:

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?

Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide family carers (FC): first interview

Introduction

XX (name patient) and you received XX months ago/ a while ago the diagnosis of ALS. Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first signals/symptoms?
- When did you notice that there was something wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you (as carer) the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

What do you know about the disease/trajectory of the disease? Which impact does ALS have?

- How did you receive this information? Who told you about this?
- What did you think/feel when you've heard this?

Do you think about your future and the future of XX (name patient)? Is it something that you worry about? Are you thinking more about the future than before the diagnosis, or vice versa, do you think about it less? Or is it another way of thinking about your future? Could you tell me something more about this?

Option 1: FC says (s)he thinks about the future:
- What do you expect? What do you hope for – for you and XX (name patient)?
- What is important for you when you think about the future? What do you think is important for the future of XX (name patient)?
- What makes you worry about the future?
- With whom would you prefer to discuss the future?

Option 2: FC thinks about the future, but they do not discuss it with the patient:
- Would you like to discuss it one day with XX (name patient)?
- Do you know what (s)he thinks about? Do you know what (s)he would want or hopes or expects about the future? What do you think is important for XX (name patient)?

Option 3: FC says they do not want to think about the future:
- You say you do not want to think about the future: How come? What makes you to refuse to think about the future?
- Is it something you are afraid of and why?

How do you think XX (name patient) thinks about it? Would (s)he thinks about the future? Would (s)he worries about it?

Thinking and talking about the future

Do you think about the care XX (name patient) might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for?

First asking the questions to see the perspective of the family carer AND then asking about how they think the patient thinks about these things.

- From whom did you receive information about his/her care?

Option 1: If FC thinks about future care:

- Is it something you discuss with XX (name patient)? Or with someone else? If so, who?
- If it is not possible to discuss care with patient/HCP:
  - Would you discuss it with friends/family/other professionals/etc?
- FC says (s)he tries to discuss it, but patient does not wish to have a conversation about his/her care:
  - How do you cope with it when you realize XX (name patient) does not want to talk about it?
  - What do you feel/think?
  - What would you like to say to XX (name patient)?
- FC indicates that they communicate openly about future care (go further to questions hereinafter).
- FC indicates they did not discuss future care, but thinks the patient might want to talk about it:
  - What do you hope this conversation would trigger? What are your expectations?
  - What would you like to say in that conversation?
  - How would that be to discuss your worries about XX (name patient) about his/her future?
  - When would it be important – according to you – to discuss it?
  - How would you experience such conversation?
  - Who should ideally start this conversation? Would you start it? Would you like that someone is there to support you? If so, who?
  - Would you prefer to have this discussion alone with XX (name patient) or together with a HCP? Why?

Option 2: FC indicates they do not wish to think about future or future care:

- Why do you not wish to think about future care?
- Does XX (name patient) share this opinion? How do you feel about the future care?
- How would you react/What would you do if XX (name patient) or a HCP wants to discuss future care and starts this conversation? Would you block it or would you talk about it? Why?

Preferences about the future/future care

What is important for you when you think about the future or future care of XX (name patient)? What do you wish for? What are your preferences/ideas? Are there things you know XX (name patient) would want or would not want about care? Are there things about care you do not want? What is – according to you important about XX's (name patient) future? What is important for you?

***Here we can give examples they have indicated during the conversation or ask how they felt about the wheelchair or walking cane etc → **ATTENTION**: listen carefully what the FC says during the interview → you cannot talk about care wishes they did not talk about. So DON'T talk about a stomach-pump if they did not talk about it): For example you might ask: how do feel about the wheelchair? How do you think XX (name patient) feels about the wheelchair? OR I have heard you've made a lot of travels before your illness, is it something you wish to do with XX (name patient)? Do you think (s)he would enjoy this? Did you discuss it? Did you make plans? ***

OPTION 1: FC knows which preferences/wishes the patient has and they seem to talk about these preferences/wishes with one another:

- I hear that you have discuss it. Was there someone else present during this conversation? If so, who?
- What have you discussed?
- What did the conversation trigger? Did you believe the moment was ideal? Why?
    - In case they did not believe the timing was ideal: When would be the best timing?
- During the conversation, have your feelings been taken into account about certain wishes/preferences/ideas/expectations/hopes/etc XX (name patient) has? Did you say how you felt/thought about it? Why and how?
    - In case not: What makes you did not say how you felt? What might happen if you did?
- Did you make concrete plans about the future/future care?
- Have you considered to write these wishes/preferences/etc down? Why (not)?
    - How do you feel about a living will?
- Imagine you need to make a medical decision instead of XX (name patient), would you believe that you can make this decision? In other words, do you know what XX (name patient) wants or does not want?
- How did the conversation end? What have you done afterwards?

Option 2: FC seems to know about the preferences/wishes the patient have about the future/future care, but they did not discuss it concrete:

- What makes you so sure that XX (give example about a preferences/wish) is what XX (name patient) wants? Have you ever discussed it? Did you ever talk about before diagnosis? Or is it something that you see/feel? Would you like to elaborate this more?
- Would you like to have a conversation about the preferences/wishes with XX (name patient)? Why (not)?

  - What would you say or do if XX (name patient) says a wish/preference you Imagine that during this conversation XX (name patient) tells a wish/preference you cannot support?
  - When would be the ideal moment for you to have this discussion?
  - With whom you would like to discuss it? Only with XX (name patient) or also with HCP/kids/friends/etc?

OPTION 3: FC indicates (s)he does not know which preferences/wishes the patient has, but would like to know them:

- I hear you wish to have a conversation about the wishes/preferences XX (name patient) might have about the future/future care. Would this be something you would like to discuss alone with XX (name patient) or with a HCP or with kids/family/friends/etc?
- Would you initiate the conversation, or would you need help?
- Is there something that would make it difficult to discuss the wishes/preferences? If so, what? And what might help to overcome this barrier?
- What are the preferences you have about the future or future care of XX (name patient)?
- What do you hope to achieve in this conversation? What are your expectations?

OPTION 4: FC indicates (s)he does not know which preferences/wishes/etc the patient has, but FC does not want to know either:

- Why do you not want to know the preferences/wishes of XX (name patient)?

What is the worst possible thing that might happen if you would discuss it? Or if you would know the wishes/preferences of XX (name patient)?

Preferences about the conversation about future/future care

OPTION 1: FCs did not discuss wishes/preferences with patient/HCP/

- When would be the best timing to have this discussion with XX (name patient)?
- What makes it difficult to start this conversation?
- How do you think this conversation might go?
- How would you feel during such conversation?

Option 2: In case they have had this discussion:

- What triggered the conversation? How did it start?
- Who was present during this conversation? Was everyone present you and XX (name patient) would have wanted?
- Did someone help you with this conversation?
- How was the timing of this conversation? Did you feel the timing was right? Why (not)?
- How did you experience this conversation?
- Was everything discussed what you and XX (name patient) wished to discuss? Would you this conversation again in the future? Why (not)?

End of interview

Has everything been discussed what you’ve had liked to discuss?

Interview patient- FC together: first interview

Introduction

XX (name patient) and you received XX months ago/ a while ago the diagnosis of ALS. Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first signals/symptoms?
- When did you notice that there was something wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you (as patient and as carer) the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

What do you know about your disease/trajectory of the disease? Which impact does ALS have?

- How did you receive this information? Who told you about this?
- What did you think/feel when you've heard this?

Do you both think about your future? Is it something that you worry about? Are you thinking more about the future than before the diagnosis, or vice versa, do you think about it less? Or is it another way of thinking about your future? Could you tell me something more about this?

Option 1: Both telling that they think about the future:
- What do you expect? What do you hope for?
- What is important for you when you think about the future?
- What makes you worry about the future?
- With whom would you prefer to discuss the future?
- Do you discuss the future together? Why (not)?

Option 2: Both think about the future, but do not discuss this:
- Would you like to discuss it one day with each other? Why (not)?
- Do you know what (s)he thinks about? Do you know what (s)he would want or hopes or expects about the future? What do you think is important?

Option 3: In case one or both does not think about the future:
- You say you do not want to think about the future: How come? What makes you to refuse to think about the future?
- Is it something you are afraid of and why?

Thinking and talking about the future

Do you think about the care you might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for? (patient)

Do you, as a carer think about the care XX (name patient) might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for? (family carer)

- From whom did you receive information about the care?

Option 1: Both think about future care:

- Is it something you discuss together? Or with someone else? If so, who?
- Both indicate that they communicate openly about future care (go further to questions hereinafter).
- Both indicate they did not discuss future care, but both think about discussing it together:
    - What do you hope this conversation would trigger? What are your expectations?
    - What would you like to say in that conversation?
    - How would that be to discuss your worries about XX (name patient) about his/her future and vice versa?
    - When would it be important – according to you both – to discuss it?
    - How would you experience such conversation?
    - Who should ideally start this conversation? Would you start it? Would you like that someone is there to support you? If so, who?
    - Would you prefer to have this discussion alone or together with a HCP? Why?

Option 2: Both indicate they do not wish to think about future or future care:

- Why do you not wish to think about future care?
- How do you feel about the future care?
- How would you react/What would you do if an HCP wants to discuss future care and starts this conversation? Would you block it or would you talk about it? Why?

Preferences about the future/future care

What is important for you when you think about the future or future care?
What do you wish for? What are your preferences/ideas? Are there things you would want or would not want about care? Are there things about care you do not want? (patient)

***Here we can give examples they have indicated during the conversation or ask how they felt about the wheelchair or walking cane etc → **ATTENTION**: listen carefully what the FC says during the interview → you cannot talk about care wishes they did not talk about. So DON'T talk about a stomach-pump if they did not talk about it): For example you might ask: how do feel about the*

*wheelchair? OR I have heard you've made a lot of travels before your illness, is it something you wish to do? Did you discuss it? Did you make plans? ***

To family carer:

- What do you think, now you hear these preferences?
- How do you feel?
- Did you know that this was important for XX (name patient)? How did you know? Did you discuss this in the past? Why (not)?

OPTION 1: Patient expresses wishes and they have talked about these preferences/wishes with one another:

- I hear that you have discuss it. Was there someone else present during this conversation? If so, who?
- What have you discussed?
- What did the conversation trigger? Did you believe the moment was ideal? Why?
    - In case they did not believe the timing was ideal: When would be the best timing?
- During the conversation, have your feelings been taken into account about certain wishes/preferences/ideas/expectations/hopes/etc XX (name patient) has? Did you say how you felt/thought about it? Why and how?
    - In case not: What makes you did not say how you felt? What might happen if you did?
- Did you make concrete plans about the future/future care?
- Have you considered to write these wishes/preferences/etc down? Why (not)?
    - How do you feel about a living will?
- Imagine you need to make a medical decision instead of XX (name patient), would you believe that you can make this decision? In other words, do you know what XX (name patient) wants or does not want?
- How did the conversation end? What have you done afterwards?

OPTION 3: FC indicates (s)he did not know which preferences/wishes the patient has, but is glad that the patient gave some wishes and preferences during the interview:

- Do you believe that you would discuss these in more depth? Why (not)? Would this be something you would like to discuss alone with XX (name patient) or with a HCP or with kids/family/friends/etc?
- Who would initiate the conversation?
- What do you hope to achieve in this conversation? What are your expectations?

OPTION 4: Both did not say any wishes or preferences, and both indicate they do not wish to discuss it:

- Why do you not want to know the preferences/wishes of XX (name patient)?
- What is the worst possible thing that might happen if you would discuss it?

Preferences about the conversation about future/future care

OPTION 1: Both did not discuss wishes/preferences with patient/HCP/

- When would be the best timing to have this discussion with XX (name patient)?
- What makes it difficult to start this conversation?
- How do you think this conversation might go?
- How would you feel during such conversation?

Option 2: In case they have had this discussion:

- What triggered the conversation? How did it start?
- Who was present during this conversation? Was everyone present you and XX (name patient) would have wanted?
- Did someone help you with this conversation?
- How was the timing of this conversation? Did you feel the timing was right? Why (not)?
- How did you experience this conversation?
- Was everything discussed what you and XX (name patient) wished to discuss? Would you this conversation again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide for patient with ALS: subsequent interviews (interview 2 and 3)

NOTE: This interview guide only shows the main questions, because the subsequent interviews build upon what has been said in the previous interview.

Introduction

Could you tell me something more about the past 3 months, since our last conversation?

Did you think about our last conversation? Do you wish to clarify something?

What were for you the biggest changes since our last conversation? (*Changes physically, emotionally, socially)*

Thinking about the future

In the previous conversation, we have discussed thinking about the future. You told me XX (what they have told)

- How do you think about it now? Did it change? Why (not)?

Thinking and talking about the future

In the previous conversation, we have discussed talking about the future and then you have indicated you did (not) discuss the future with XX (name family carer) or with someone else

- Did you have this discussion in the past months? Why (not)?

OPTION 1: Patient did not talk about the future, and do not wish to talk about it:

- What makes it difficult to think about the future?
- Does XX (name FC) has the same feeling?
- How would you react when XX (name FC) starts the conversation? Would you block it? Why (not)?
- What are you afraid of that might happen if you talk about this?

OPTION 2: Patient thinks about the future, and wish to talk about the future, but family carer blocks the conversation:

- How do you feel about this?
- What do you do when that happens?
- What would you like to tell him/her?

OPTION 3: Patient thinks about the future, and has discussed it

- What triggered the conversation?

- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

- **NOTE:** ask how the previous interview influenced the possible conversation

Preferences about the future/future care

In the previous interview, we have discussed preferences about the future/future care. You said following things XX

- Did wishes/preferences/ideas change during these past months? Why (not)?
- In case wishes/preferences/ideas have changed
    - What triggered the change in your wishes/preferences/ideas?
    - Did you discuss these changes?
        - If so, with whom?
        - If not, why not?
- **NOTE:** ask how the previous interview influenced the possible change

In the previous interview, you told me that:

OPTION 1: you would want to discuss your preferences in the near future:

- Have you done this in the past months? Why (not)?
- In case family carers blocks the discussion:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?

OPTION 2: you have discussed your preferences prior to the first interview with XX (the persons they have indicated in the previous interview to whom they have discussed it)

- Have you discussed it again in the past months? Why (not)?

OPTION 3: I hear you have discussed your preferences in the past months

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

OPTION 4: you have not discussed your preferences and you do not wish to discuss this

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

In the previous conversation you have told me that the ideal timing for this conversation would be XX

OPTION 1: the ideal timing has arrived, but patient did not have this conversation

- Why did you not have this conversation? What stopped you?

OPTION 2: the ideal timing did not arrive, and there was no discussion in the meantime:

- Do you still have the same idea about the ideal timing to have this conversation? Why (not)?

OPTION 3: the ideal timing did not arrive, but patient did have a discussion in the past months:

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide for FC: subsequent interviews (interview 2 and 3)

NOTE: This interview guide only shows the main questions because the subsequent interviews build upon what has been said in the previous interview.

Introduction

Could you tell me something more about the past 3 months, since our last conversation?

Did you think about our last conversation? Do you wish to clarify something?

What were for you the biggest changes since our last conversation? (*Changes physically, emotionally, socially)*

Thinking about the future

In the previous conversation, we have discussed thinking about the future. You told me XX (what they have told)

- How do you think about it now? Did it change? Why (not)?

Thinking and talking about the future

In the previous conversation, we have discussed talking about the future and then you have indicated you did (not) discuss the future with XX (name patient) or with someone else

- Did you have this discussion in the past months? Why (not)?

OPTION 1: FC did not talk about the future, and do not wish to talk about it:
- What makes it difficult to think about the future?
- Does XX (name patient) has the same feeling?
- How would you react when XX (name patient) starts the conversation? Would you block it? Why (not)?
- What are you afraid of that might happen if you talk about this?

OPTION 2: FC thinks about the future, and wish to talk about the future, but patient blocks the conversation:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?

OPTION 3: FC thinks about the future, and has discussed it
- What triggered the conversation?

- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

- **NOTE:** ask how the previous interview influenced the possible conversation

Preferences about the future/future care

In the previous interview, we have discussed preferences about the future/future care. You said following things XX

- Do you believe the wishes/preferences/ideas of XX (name patient) has changed during these past months? Why (not)?
- In case wishes/preferences/ideas have changed
    - What triggered the change in their wishes/preferences/ideas?
- **NOTE:** ask how the previous interview influenced the possible change

In the previous interview, you told me that:

OPTION 1: you would want to discuss the preferences with XX (name patient) in the near future:

- Have you done this in the past months? Why (not)?
- In case patient blocks the discussion:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?

OPTION 2: you have discussed with XX (name patient) the preferences prior to the first interview

- Have you discussed it again in the past months? Why (not)?

OPTION 3: I hear you have discussed the preferences with XX (name patient) in the past months

- What triggered the conversation?
- Who was present?
- Who initiated the conversation? You or XX (name patient) or someone else?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

OPTION 4: you have not discussed the preferences with XX (name patient) and you do not wish to discuss this

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

In the previous conversation you have told me that the ideal timing for this conversation would be XX

OPTION 1: the ideal timing has arrived, but FC did not have this conversation

- Why did you not have this conversation? What stopped you?

OPTION 2: the ideal timing did not arrive, and there was no discussion in the meantime:

- Do you still have the same idea about the ideal timing to have this conversation? Why (not)?

OPTION 3: the ideal timing did not arrive, but FC did have a discussion in the past months:

- What triggered the conversation?
- Who was present?
- Who initiated the conversation? You, XX (name patient) or someone else?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you’ve had liked to discuss?