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Research priorities relating to the debate on assisted dying: what do we still need to know? Results of a modified Delphi technique

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Keywords: Assisted dying; Delphi technique; research priorities

Abstract

Objective

To identify the main areas of uncertainty and subsequent research priorities to inform the ongoing debate around assisted dying.

Design

Two-round electronic modified Delphi consultation with experts and interested bodies.

Setting and participants

110 groups and individuals interested in the subject of end of life care and/or assisted dying were approached to participate. Respondents included health and social care professionals, researchers, campaigners, patients, and carers.

In the first round, the respondents were asked to propose high-priority research questions related to the topic of assisted dying. The collected research questions were then de-duplicated and presented to all respondents in a second round in which they could rate each question in terms of importance.

Results

24% and 26% of participants responded to the first and second rounds respectively. Respondents suggested 85 unique research questions in the first round. These were grouped by theme and rated in terms of importance in the second round.

Emergent themes were: Palliative care/symptom control; patient characteristics, experiences and decisions; families and carers; society and the general public; arguments for and against assisted dying; international experiences /analysis of existing national data; suicide; mental health, psychological and psychosocial considerations; comorbidities; the role of clinicians; environment and external influences; broader topics incorporating assisted dying; and moral, ethical and legal issues.

Ten of the 85 proposed questions were rated as being important ($\geq 7/10$) by at least 50% of respondents.

Conclusions

Research questions with the highest levels of consensus were predominately concerned with understanding how and why people make end-of-life decisions, and which factors influence those decisions. Dissemination of these findings alongside a focused examination of the existing literature may be the most effective way to add evidence to the ongoing debate around assisted dying.

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Strengths and weaknesses

- To our knowledge, this is currently the only attempt to identify the main areas of uncertainty and subsequent research priorities to inform the ongoing debate around assisted dying.
- All research questions were generated and refined by a group of interested parties from both sides of the debate, most of whom considered themselves to be very knowledgeable about the topic.
- The research priorities were collectively identified by health and social care professionals, researchers, campaigners, patients and carers, rather than by researchers alone.
- The response rate and overall level of consensus as defined in this study were relatively low.
- Very few of the religious groups that we approached responded to the survey.

Introduction

Public debate around end of life issues has increased in recent years, partly because of demographic changes caused by a rapidly aging population without a corresponding increase in healthy life expectancy.¹ The debate on assisted dying has become particularly prominent, to the extent that individual 'right to die' appeals frequently receive national media coverage²⁻⁴ and the topic has even been explored in popular soap opera storylines.⁵

'Assisted dying' is not a legal term, but is typically understood to mean a circumstance in which a chronically or terminally ill person is allowed to end their own life, either by assisted suicide (the patient is given lethal drugs to take themselves) or euthanasia (somebody else administers lethal drugs to the patient).

The majority of jurisdictions that have legalized assisted dying permit assisted suicide (in which the patient must themselves take action to end their own life), though some permit voluntary euthanasia or both. Some form of assisted dying is legal in Columbia, Switzerland, Netherlands, Belgium, Luxembourg, Canada, and the US states of Oregon, Washington, Montana and Vermont.

Assisted dying remains illegal in the UK, with proposed bills for legalization recently having been strongly rejected by both MSPs and MPs in May 2015 and September 2015 respectively.^{6,7} Contrastingly, legislators in the state of California have agreed to allow doctor-assisted dying from June 2016.⁸

The debate around any proposed legal changes is strongly polarized, and a number of opposing arguments have been put forward. Many arguments focus on moral, ethical, religious or legal issues and are often strongly informed by pre-existing beliefs. Far fewer arguments are based on objective empirical evidence, or else evidence is used only selectively. As such, debates often end in impasse.

Some key areas of debate include: individual autonomy, patient choice, the roles of doctors and relatives in assisting death, the nature of 'unbearable' illness, the impact of availability and efficacy of palliative care, the effectiveness of safeguards to protect vulnerable people, and the extent of 'unseen' assisted dying/suicide within the current system. Each of these raises questions about "what we know" about the current state of affairs, whether there is a need for further objective

evidence to illuminate the debate on assisted dying, and if so, what focus and form this evidence might take.

We therefore conducted a modified Delphi exercise with experts and interested parties to identify the main areas of uncertainty and subsequent research priorities to inform the ongoing debate around assisted dying.

Method

Design

The Delphi technique is a structured and iterative method for collecting anonymous individual opinions from a panel of topic experts where a consensus is required. The basic principle is for the panel to receive successive questionnaires, each one containing the anonymous responses to the previous round, and for them to modify their responses until a consensus is reached.⁹⁻¹¹ The method has been used to identify research priorities in a number of different topic areas.¹²⁻¹⁴

For the debate around assisted dying, this approach allowed respondents to generate a number of relevant research questions and then identify those of the highest priority. The aim was to identify any commonly agreed areas of perceived uncertainty, regardless of prior stance or beliefs.

In the first of two rounds, participants were invited to suggest areas of uncertainty that could be addressed by research:

“A number of arguments have been made both for and against changing the law around assisted dying. Some of these are moral or ethical arguments, others are legal arguments and others are medical or pragmatic arguments. These different arguments require differing degrees of supporting evidence. The aim of this study is to identify the most important areas where the facts are unknown or uncertain, either because there is no evidence or because the evidence we have is limited.

In this first stage, we would simply like you to suggest - as concisely as possible - where you think there is a need for either new evidence or a better summary of the existing evidence in this area.”

Participants were presented with examples of research questions and then provided space to suggest their own (see Appendix). No restrictions were placed on the number of suggestions a participant could make. After the first round was closed, thematic analysis was used to de-duplicate, code, and group items.¹⁵

In the second round, participants were asked to rate the importance of each research question on a scale from 1 (not at all important) to 10 (extremely important). Participants were encouraged to give a low score to questions that have already been fully answered or are of little interest, but to highly score questions where there is still uncertainty and where research is urgently needed. Questions receiving a score of ≥ 7 points on the ten-point scale were interpreted as having high importance. A ‘No opinion’ option was also provided. Since the respondent population in this second round was unlikely to be identical to that in the first round, participants in both stages were briefly asked to rate their current level of knowledge and give their stance on hypothetical changes in the law.

Results of both rounds were shared with respondents.

Questionnaires were administered electronically using on-line survey software Survey Monkey (www.surveymonkey.com).

Participants

Experts and parties interested in the subject of end of life care and/or assisted dying (clinicians, charities, religious groups, think-tanks, pressure groups, patient and carer representatives) were approached to participate. All were based in Europe, predominately in the UK.

An initial list of individuals, organisations and groups was identified through searches of both academic, government and mass media publications, including the 2012 report published by the Commission on Assisted Dying in which a broad range of interested parties submitted evidence.¹⁶ In addition, pro- and anti-assisted dying groups (e.g. Dignity in Dying, Living and Dying Well, Care Not Killing) were contacted directly.

Email addresses were collected from personal contact lists and publicly available sources (e.g., organisational websites). All emails were personalised to individuals. Groups were encouraged to disseminate the invitation to their members via their own websites, electronic mailing lists and newsletters. Anyone responding to a link cascaded by an original contact was added to the contact list and sent a questionnaire. All contacts were assured confidentiality, with the aim of encouraging participation and openness.

All contacts were invited to both rounds of the survey, including first-round non-responders (unless they had chosen the option to withdraw from further contact).

In order to assess representation from different stakeholder groups and identify any differences in the responses between them, participants were also asked to categorise and/or briefly describe their background, rate their self-perceived level of knowledge on the topic of assisted dying, and give their general prior opinion on the issues of assisted dying and voluntary euthanasia.

Backgrounds were categorized as: clinician, patient, carer/ex-carer, patient representative, researcher, campaigner, legislator, or other. Respondents could choose one or more categories and were encouraged to provide further details where necessary.

Respondents' prior opinions were elicited with three questions on hypothetical changes to the existing law. These asked: "Do you think that under certain defined circumstances, the law should allow..."

1. ...assisted suicide (providing someone with the means to end his or her own life)?
2. ...voluntary euthanasia (ending another person's life at their own request) by a doctor?
3. ...voluntary euthanasia (ending another person's life at their own request) by a close family member?

These three scenarios reflect proposed or existing legal arrangements in jurisdictions outside the UK and reflect the chief options contested in the debate around assisted dying. Respondents could reply "Yes", "No", "Don't know", or "Rather not say".

Questionnaires

Questionnaires were piloted before distribution, resulting in minor wording changes to improve clarity. Round one was initially 'open' for responses for two weeks, though this was extended by a week after several contacts requested extra time to formulate their questions. Round two was open for three weeks. Reminder emails were sent to all contacts approximately one week before the close of each round. A mixture of pre-specified and free text responses were used to facilitate ease of response and analysis of data. In order to ensure that sufficient data were collected and to prevent inadvertent missing data, 'pick list' questions were made mandatory. It was not mandatory to complete the free text boxes.

Analysis

All responses were collected in 'Survey Monkey' for initial tabulation and analysis. Subsequent analyses and outputs were produced in Excel. Descriptive statistics were calculated and used to investigate the distribution of scores. An initial consensus level was set at a score of ≥ 7 points on the ten-point importance scale from $\geq 50\%$ of respondents. Where a respondent did not provide a score, this value was recorded as missing; there was no imputation of missing values.

The relationship between respondents' prior position and scoring of higher consensus questions was explored. Variances were unequal across groups, with small numbers of observations for some questions, precluding the calculation of meaningful hypothesis tests. Therefore these data were displayed graphically.

Ethical approval

Invitees were promised anonymity and submission of completed questionnaires was taken as implied consent. The research was approved by the University of York Health Sciences Research Governance Committee.¹⁷

Results

A total of 110 individuals and organizations were initially invited to participate. An additional 16 contacts were suggested by first round respondents, and a further 31 were suggested by second round respondents, all of whom were also invited to participate.

First round

Respondent characteristics

30 (24%) invitees (28 of whom completed all questions) responded to the initial questionnaire that required the formulation of research questions. Seven invitees declined the invitation, one email was undeliverable, and the remainder provided no response.

75% of respondents rated themselves as being "very knowledgeable" or "expert" on the topic of assisted dying (Table 1). Only one respondent considered themselves to have "limited knowledge".

Respondent backgrounds included health and social care professionals (palliative care, oncology, nursing, general practice, public health, social work), researchers, campaigners, and patients or their carers/representatives (Table 2).

Opinions on assisted dying and voluntary euthanasia were mixed. In response to the question “Do you think that under certain defined circumstances, the law should allow assisted suicide (providing someone with the means to end his or her own life)?”, 50% of respondents answered “Yes”, 36% answered “No” and 14% responded that they did not know or would rather not say. A similar pattern of responses was seen for voluntary euthanasia by a doctor (Yes 46%, No 40%, Don’t know/Rather not say 14%), though a majority of respondents were opposed to voluntary euthanasia by a close family member (60% vs. 21% in favour and 18% uncertain or unwilling to respond).

Table 1: Respondents' self-rated knowledge

How would you rate your own knowledge on the topic of assisted dying?	Round 1		Round 2	
	Count	%	Count	%
Know nothing	0	0	1	2.9
Limited knowledge	1	3.6	2	5.7
Reasonably knowledgeable	6	21.4	14	40.0
Very knowledgeable	15	53.6	12	34.3
Expert	6	21.4	6	17.1
TOTAL	28	100	35	100

Table 2: Respondents' background

How would you describe your background?	Round 1	Round 2
Clinician	11	9
Patient	2	2
Carer or ex-carer	1	9
Patient representative	5	4
Researcher	8	9
Campaigner	5	9
Legislator	0	0
Other	5	5
Additional background details (where reported)		
Palliative care	4	5
Hospital consultant	4	1
Oncology	1	1
Public health	1	0
Gerontology	0	1
Nursing	1	0
General practice	1	2
Psychiatry	0	1
Right to die group/organisation	3	2
Policy advice	2	0
Social work	1	1
Charity sector	2	1
Law	0	1
Chaplaincy	0	1
Veterinary medicine	0	1

Relative	0	1
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Respondent-generated research questions and themes

Respondents made 102 separate suggestions for research questions. Thematic analysis identified thirteen interrelated themes (Table 3)

Table 3: Research question themes

Theme	Number of suggested questions
Palliative care/symptom control	7
Patient characteristics, experiences and decisions	13
Families and carers	3
Society and the general public	7
Arguments for and against assisted dying	4
International experiences / analysis of existing national data	18
Suicide	2
Mental health, psychological and psychosocial considerations	9
Comorbidities	3
The role of clinicians	7
Environment and external influences	4
Broader topics incorporating assisted dying	2
Moral, ethical and legal issues	6

Where two or more responses expressed an identical research question in different ways, the most concise formulation was kept. Where two or more responses expressed similar but not necessarily identical questions, both versions were retained for the second round. Some responses were categorized as “Non research questions” where they posed largely moral/ethical questions or statements of opinion that could not be rephrased without the risk of changing the meaning intended by the respondent (e.g. “Moral arguments around autonomy and dignity”).

After de-duplication, a total of 85 questions in the 13 categories were sent to participants to be rated in the second round (see Appendix).

Second round

Respondent characteristics

A greater number of participants (n=39; 26%) responded in the second round than the first. Respondent backgrounds, where reported, were similar between rounds, though a greater number of respondents in the second round categorized their backgrounds as “carer / ex-carer”, “campaigner” or both (Table 2).

Participants were again asked to rate their current level of knowledge and give their stance on hypothetical changes in the law. Compared with the first round, a smaller proportion of respondents (51.4%) rated themselves as being “very knowledgeable” or “expert” on the topic of assisted dying than did in the first round (Table 1).

A greater proportion of respondents were sympathetic to assisted dying by a doctor than in the first round (74% vs 50% and 66% vs 46% opposed to voluntary euthanasia by a close family member (54%; List of Figures Figure).

Respondent ratings of research questions

A median of 32 ratings (range 23 to 36) were available for each of the 85 questions.

The distribution of scores was negatively skewed (i.e. in favour of high ratings of importance) for all questions, but varied widely between individual respondents (76 questions received ratings ranging from 1 to 10 points). Appendix shows box-and-whisker plots summarizing the mean, median, range and interquartile range values for each question.

The level of consensus among respondents on whether a question was important (i.e. score ≥ 7) ranged from 4% to 56% (see appendix for details)

Summary of highest priority questions

Ten of the 85 rated questions met the consensus level of receiving a score of ≥ 7 points from $\geq 50\%$ of respondents. These are shown in Table 4.

Table 4: Questions rated $\geq 7/10$ by at least 50% of respondents

Primary theme	Proposed research question	Consensus
Families and carers	What are the effects of carer burden on requests for assisted dying?	55.9%
Arguments for and against assisted dying	Understanding better why some patient groups are strongly opposed to assisted suicide - what are their concerns, could these be mitigated?	54.3%
Comorbidities	Given the progression of dementia, when should end of life care be discussed with the person with dementia and who should initiate this discussion?	53.3%
Mental health, psychological and psychosocial issues	What are the triggers for requesting assisted dying?	52.9%
Personal characteristics, experiences and decisions	How do the views of people considering euthanasia/physician assisted suicide with a spectrum of conditions develop over time - especially those not near the end of life?	52.8%
International experiences / analysis of existing data	Does international experience confirm or lay to rest concerns that vulnerable individuals will be pressurized to avail themselves of assisted dying?	51.5%
Personal characteristics, experiences and decisions	Why do people consider going to Dignitas - is it fear of dying, pain, control?	50%
Personal characteristics, experiences and decisions	What would enhance a person's quality of life after diagnosis of a terminal illness, how do they define 'quality of life' and what are factors they take into consideration in assessing it?	50%
Broader topics	How to operationalise concepts such as "unbearable suffering"?	50%
Moral, ethical and legal issues	The best alternative care pathways for 'end of life' (rather than depriving the patient food and drink and allowing them to starve)?	50%

The question with the single highest level of consensus was:

“What are the effects of carer burden on requests for assisted dying?” (55.9%)

This was one of several highest-level consensus questions specifically concerned with understanding how and why people make end-of-life decisions, and which factors influence those decisions:

“How do the views of people considering euthanasia/physician assisted suicide with a spectrum of conditions develop over time - especially those not near the end of life?” (52.8%)

“What are the triggers for requesting assisted dying?” (52.9%)

“Why do people consider going to Dignitas - is it fear of dying, pain, control?” (50%)

What would enhance a person’s quality of life after diagnosis of a terminal illness, how do they define 'quality of life' and what are factors they take into consideration in assessing it” (50%)

These questions imply a need for robust qualitative evidence about individuals’ motivations, experiences and decisions, while another question identified the analysis of existing evidence from jurisdictions in which some form of assisted dying is already legal as an area of importance:

“Does international experience confirm or lay to rest concerns that vulnerable individuals will be pressurized to avail themselves of assisted dying?” (51.5%)

One question raised a general methodological issue for end-of-life researchers:

How to operationalise concepts such as "unbearable suffering"?” (50%)

Two consensus items touched on issues broader than simply obtaining good research evidence, raising questions about optimal planning and decision-making at the end of life:

“Given the progression of dementia, when end of life care should be discussed with the person with dementia and who should initiate this discussion?” (53.3%)

“What are the best alternative care pathways for 'end of life' (rather than depriving the patient food and drink and allowing them to starve)?” (50%)

Finally, one proposed research question was not concerned with understanding people at the end of life, but those who take a particular position on the issue of assisted dying:

“Understanding better why some patient groups are strongly opposed to assisted suicide - what are their concerns, could these be mitigated?” (54.3%)

Relationship between respondents’ views on assisted dying and their perceived importance of research questions

Figure ,

Figure and Figure illustrate the relationship between respondent views on various forms of assisted dying and mean ratings for questions with $\geq 50\%$ consensus. The pattern of scores in

Figure suggests that respondents with no fixed position on assisted suicide (i.e. "Don't know") consistently gave higher ratings to the research questions than did respondents with a fixed position (i.e. "Yes" or "No"). As might be expected, respondents with less certainty tended to value research evidence more highly.

The pattern of ratings was not consistent for all views on assisted dying (assisted suicide, doctor-assisted voluntary euthanasia, family-assisted euthanasia), though mean ratings tended to be slightly lower among respondents who were against any hypothetical changes in the law (

Figure ,

Figure Figure).

Discussion

To our knowledge, this is currently the only attempt to identify the main areas of uncertainty and subsequent research priorities to inform the ongoing debate around assisted dying. All research questions were generated and refined by a group of interested parties from both sides of the debate, most of whom considered themselves to be very knowledgeable about the topic.

New research ideas are frequently generated by research professionals who have an interest in a topic area. While researchers may be able to identify gaps in the evidence, they may not be best placed to determine which areas are most urgently in need of further research. Approaches such as the James Lind Priority Setting Partnerships aim to tackle treatment uncertainties by consulting a wider range of participants in order to identify research priorities. Such partnerships consist of at least one patient/carer organisation and at least one clinician organisation.¹⁸

This Delphi consultation incorporated a diverse set of experiences, including those of health and social care professionals, researchers, campaigners, patients and carers from a predominately UK setting. Consequently, the research priorities identified here may more closely reflect those of value to wider society. For example, researchers might be interested in analyzing data collected in jurisdictions where assisted dying is legal. However, while research questions about cross-country comparisons were most commonly suggested by panel members, only one of these emerged among the highest-priority questions based on consensus score.

The results raised a number of important questions about end-of-life issues that were broader than just the topic of assisted dying. For example, clear definition and measurement of concepts such as 'quality of life' and 'unbearable suffering' in this setting are fundamental to understanding end-of-life issues, yet the panel of experts considered the evidence to be lacking in this area. The NIHR 'Palliative and end of life care Priority Setting Partnership' (PeolcPSP) has looked at end of life issues more broadly and identified a set of unanswered questions around provision and access to palliative care and the benefits of Advance Care Planning that complement those identified in the current exercise.¹⁹ Some issues - such as concerns about how to listen to and incorporate patient preferences - overlap with the priorities identified here. Interestingly, the PeolcPSP also received a number of comments and questions outside its intended scope.

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3 The overall level of consensus as defined in this study was relatively low (4% to 56%). However, wide
4 variation in second round scores between items suggested that respondents were able to distinguish
5 pertinent research questions from untestable hypotheses and statements of opinion. Therefore it
6 seems that most respondents understood the aim of the project, and were focused on identifying
7 areas of uncertainty that would benefit from empirical investigation. Further, it seems that the
8 consensus threshold applied here (at least half the respondents giving ≥ 7 points) was able to identify
9 the highest priority questions.
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12 The response rate to the Delphi (around 25%) was relatively low, but compares favorably to other
13 surveys that have recruited doctors.^{20, 21} A low response might have been expected, given the
14 onus on respondents to formulate their own research questions; the level of time and effort
15 required for this may have been a barrier for participants who might otherwise have responded to a
16 simple 'tick-box' questionnaire. However, a fundamental objective of this process was to obtain
17 research questions from experts and other interested parties, rather than have them imposed by
18 researchers. Similar future surveys should consider engaging participants as early in the process as
19 possible, and attempt to sustain participant enthusiasm to overcome such barriers. Alternatively,
20 questions might be initially generated through interviews or focus groups.
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24 Whereas large sample sizes are important for questions of precision, the aim of the Delphi exercise
25 was to identify consensus among a diverse group of interested individuals. In this case, obtaining an
26 appropriate sampling frame is perhaps more important. As well as including participants from a
27 variety of professions, we approached groups and individuals with opposing opinions on the subject
28 of assisted dying. While respondents from both sides of the debate contributed to both stages of the
29 survey, there was a slight predominance of respondents in favour of assisted dying. Very few of the
30 religious groups that we approached responded to the survey, so this may have had an influence on
31 the ratings (i.e. through underrepresentation of anti-assisted dying opinion), and might partly
32 explain high importance ratings for the question "Understanding better why some patient groups
33 are strongly opposed to assisted suicide - what are their concerns, could these be mitigated?".
34 However, the other highly-rated questions do not appear to have an overtly pro- or anti-assisted
35 dying stance, but rather address important areas of uncertainty. The distribution of scores did not
36 show a clear influence of prior stance on question ratings: a slight tendency for lower scores among
37 those respondents against a change in the law was observed, though for most questions the scores
38 did not differ greatly. It is plausible that people who consider the current legal position to be
39 adequate are less likely to believe there is a need for research than people who are unsure or favour
40 a change in the law.
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46 **Recommendations for future research**

47 The logical next step would be to address the priority questions identified from this process. This
48 should be done in the first instance by examining the existing evidence to further refine the design
49 of any future research. There is some available evidence relevant to some of the questions identified
50 here (for example, on views of patients and carers),²² but a number of the questions have not been
51 addressed directly or systematically. Before undertaking any new primary research, one or more
52 systematic reviews of the existing evidence focusing on the themes and questions identified here
53 may be worthwhile. For example, a review of qualitative evidence specifically concerned with the
54 influence of dementia on patient and carer views related to assisted dying would be of value.
55 Though lower priority, this may be supplemented by a systematic review of the international
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evidence to determine the fate of vulnerable people in jurisdictions with legalized forms of assisted dying, which remains an area of major contention. A well conducted systematic review could provide an impartial and comprehensive overview of the evidence, making explicit its relative strengths and weaknesses in relation to the well-worn arguments in this area. If uncertainties still remain, the review could make clear and precise recommendations about where new primary research is needed.

Conclusion

This consultation revealed a number of important uncertainties around the debate on assisted dying and end-of-life issues more broadly. Eighty-five unique research questions were suggested by a broad range of interested parties with high levels of topic expertise. Research questions with the highest levels of consensus were predominately concerned with understanding how and why people make end-of-life decisions, and which factors influence those decisions. Dissemination of these findings alongside a focused examination of the existing literature may be the most effective way to bring objective research evidence into the ongoing debate around assisted dying.

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Contributorship statement

Mark Rodgers contributed to the design of the study, collected, analysed and interpreted the data, drafted, revised and approved the manuscript.

Alison Booth contributed to the design of the study, interpreted the data, and revised and approved the manuscript.

Gill Norman contributed to the design of the study, interpreted the data, and revised and approved the manuscript.

Amanda Sowden contributed to the design of the study, interpreted the data, and revised and approved the manuscript.

Competing interests

The authors have no competing interests to declare

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No additional data available

References

1. Cracknell R. *The ageing population: Key issues for the 2010 Parliament*: House of Commons Library Research; 2010. Available from: <http://www.parliament.uk/business/publications/research/key-issues-for-the-new-parliament/value-for-money-in-public-services/the-ageing-population/>
2. BBC News. Husband speaks out on wife's assisted suicide. [serial online] 2012. [cited Available from: <http://www.bbc.co.uk/news/uk-england-suffolk-17550982>
3. BBC News. *Right-to-die man Tony Nicklinson dead after refusing food*. 2012. [cited Available from: <http://www.bbc.co.uk/news/uk-england-19341722>.
4. ITV News. *Hope remains despite Supreme Court rejection of right to die campaign*. 2014. [cited Available from: <http://www.itv.com/news/calendar/2014-06-25/hope-remains-despite-supreme-court-rejection-of-right-to-die-campaign/>.
5. ITV News. *Coronation Street shows controversial right to die scene*. 2014. [cited Available from: <http://www.itv.com/news/2014-01-20/coronation-street-controversial-assisted-suicide-scene-hayley-cropper/>.
6. *Assisted Suicide (Scotland) Bill*. The Scottish Parliament; [cited 2016 7th April]. Available from: <http://www.scottish.parliament.uk/parliamentarybusiness/Bills/69604.aspx>.
7. *Assisted Dying (No.2) Bill 2015-16*. UK Parliament; [cited 2016 7th April]. Available from: <http://services.parliament.uk/bills/2015-16/assisteddyingno2.html>.
8. Clodfelter R, Adashi E. The Liberty to Die: California Enacts Physician Aid-in-Dying Law. *JAMA: Journal of the American Medical Association* 2016;315:251-2. Available from: <http://jama.jamanetwork.com/article.aspx?articleid=2482335>
9. Hsu C, Sandford B. The Delphi Technique: Making Sense of Consensus. *Practical Assessment Research & Evaluation* 2007;12:1-8.
10. Murphy E, Black N, Lamping D, McKee C, Sanderson C. Consensus development methods, and their use in clinical guideline development: a review. *Health Technology Assessment* 1998;2:1-88.
11. Sinha I, Smyth R, Williamson P. Using the delphi technique to determine which outcomes to measure in clinical trials: recommendations for the future based on a systematic review of existing studies. *PLoS Medicine* 2011;8:e1000393.
12. Cresswell K, Panesar S, Salvilla S, Carson-Stevens A, Larizgoitia I. Global Research Priorities to Better Understand the Burden of Iatrogenic Harm in Primary Care: An International Delphi Exercise. *PLoS Medicine* 2013;10:e1001554.
13. Rankin G, Rushton A, Olver P, Moore A. Chartered Society of Physiotherapy's identification of national research priorities for physiotherapy using a modified Delphi technique. *Physiotherapy* 2012;98:260-72.

- 1
2
3 14. Wielenga J, Tume L, Latour J, van den Hoogen A. European neonatal intensive care nursing
4 research priorities: an e-Delphi study. *Archives of Disease in Childhood-Fetal and Neonatal Edition*
5 2015;100:F66-71.
6
7 15. Guest G, MacQueen K, Namey E. *Applied thematic analysis*. Thousand Oaks: SAGE; 2011.
8
9 16. The Commission on Assisted Dying. 2011. [cited Available from:
10 [http://www.demos.co.uk/files/476_CoAD_FinalReport_158x240_I_web_single-](http://www.demos.co.uk/files/476_CoAD_FinalReport_158x240_I_web_single-NEW_.pdf?1328113363)
11 [NEW_.pdf?1328113363](http://www.demos.co.uk/files/476_CoAD_FinalReport_158x240_I_web_single-NEW_.pdf?1328113363).
12
13 17. University of York Health Sciences Research Governance Committee. 2015. [cited Available from:
14 <http://www.york.ac.uk/healthsciences/research-information/rsg/>.
15
16 18. James Lind Alliance. *Priority Setting Partnerships*. 2015. [cited Available from:
17 <http://www.lindalliance.org/Priority%20Setting%20Members.asp>.
18
19 19. Palliative and end of life care Priority Setting Partnership. *Putting patients, carers and clinicians*
20 *at the heart of palliative and end of life care research*. 2015. [cited Available from:
21 http://www.palliativecarepsp.org.uk/wp-content/uploads/2015/01/PeolcPSP_Final_Report.pdf.
22
23 20. Baruch Y. Response rate in academic studies-A comparative analysis. *Human Relations*
24 1999;52:421-38.
25
26 21. Scott A, Jeon S, Joyce C, Humphreys J, Kalb G, Witt J. A randomised trial and economic evaluation
27 of the effect of response mode on response rate, response bias, and item non-response in a survey
28 of doctors. *BMC Medical Research Methodology* 2011;11:126.
29
30 22. Hendry M, Pasterfield D, Lewis R, Carter B, Hodgson D, Wilkinson C, et al. Why do we want the
31 right to die? A systematic review of the international literature on the views of patients, carers and
32 the public on assisted dying. *Palliative medicine* 2013;27:13-26.
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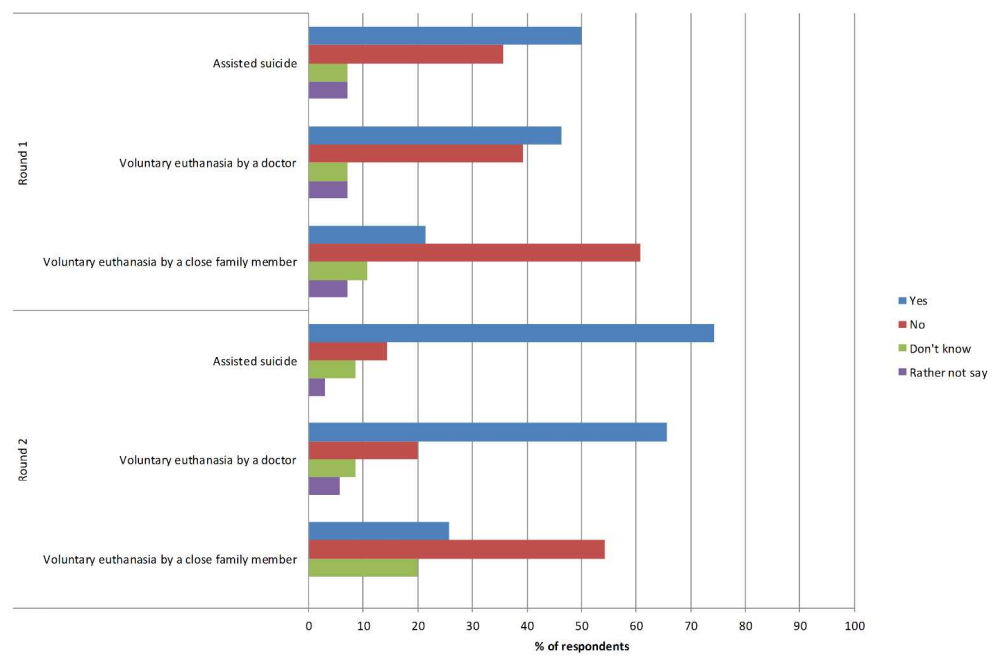


Figure 1: Respondent support for a change in the law
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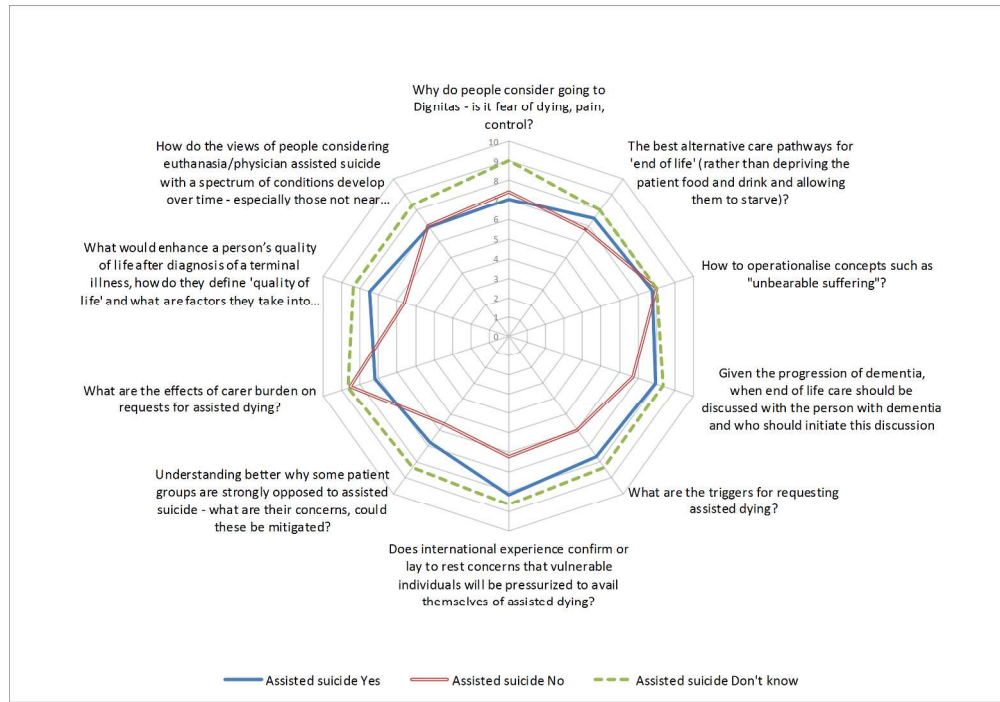


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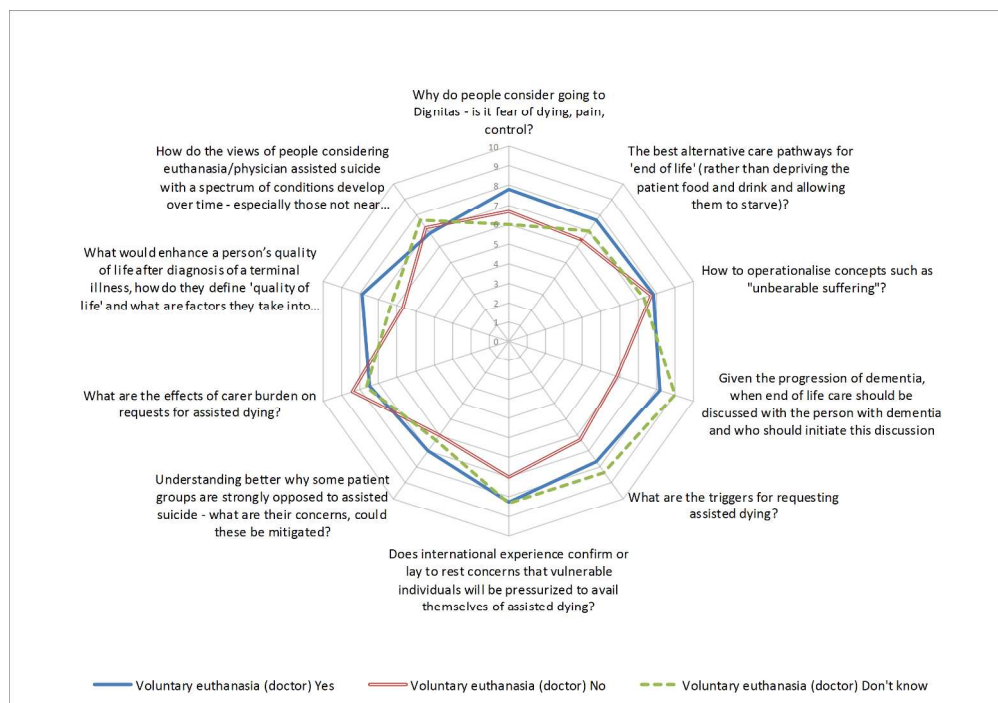


Figure 3: Relationship between respondent views on doctor-assisted voluntary euthanasia and mean rating for highest consensus questions
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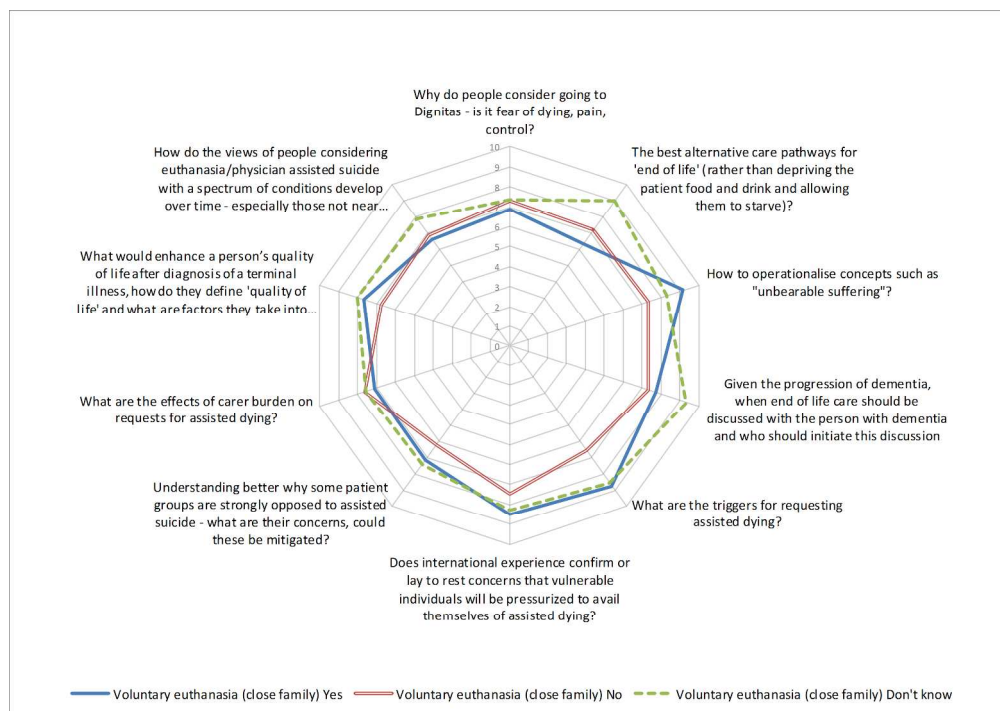


Figure 4: Relationship between respondent views on family-assisted voluntary euthanasia and mean rating for highest consensus questions
245x172mm (300 x 300 DPI)

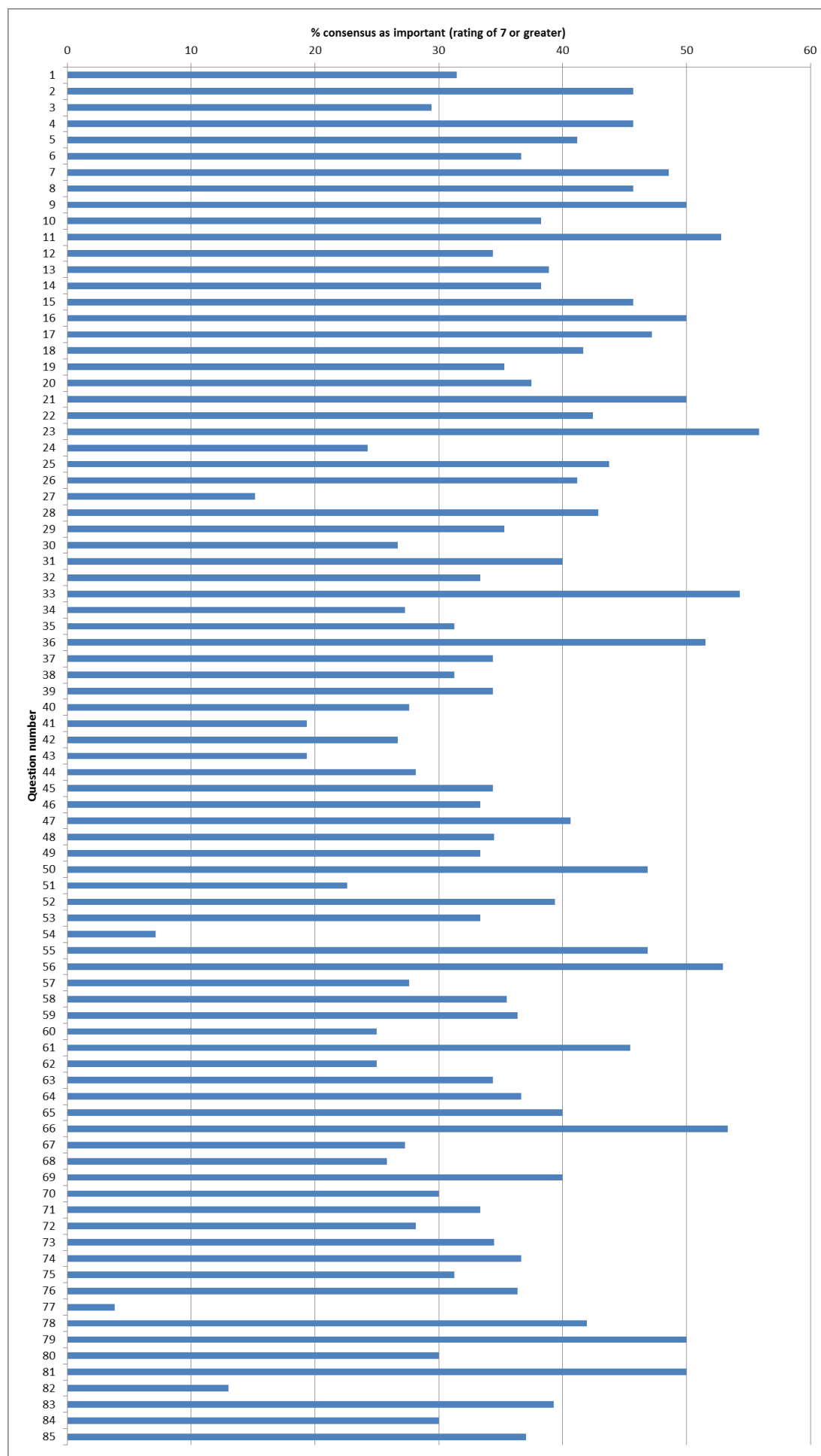
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Appendix 1: Box and whisker plots of all responses to questions rated in round 2



Appendix 2: Percentage consensus on importance of questions listed in round 2



BMJ Open

Research priorities relating to the debate on assisted dying: what do we still need to know? Results of a modified Delphi technique

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Research priorities relating to the debate on assisted dying: what do we still need to know? Results of a modified Delphi technique

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Keywords: Assisted dying; Delphi technique; research priorities

Abstract

Objective

To identify the main areas of uncertainty and subsequent research priorities to inform the ongoing debate around assisted dying.

Design

Two-round electronic modified Delphi consultation with experts and interested bodies.

Setting and participants

110 groups and individuals interested in the subject of end of life care and/or assisted dying were approached to participate. Respondents included health and social care professionals, researchers, campaigners, patients, and carers predominately based in the UK.

In the first round, the respondents were asked to propose high-priority research questions related to the topic of assisted dying. The collected research questions were then de-duplicated and presented to all respondents in a second round in which they could rate each question in terms of importance.

Results

24% and 26% of participants responded to the first and second rounds respectively. Respondents suggested 85 unique research questions in the first round. These were grouped by theme and rated in terms of importance in the second round.

Emergent themes were: Palliative care/symptom control; patient characteristics, experiences and decisions; families and carers; society and the general public; arguments for and against assisted dying; international experiences /analysis of existing national data; suicide; mental health, psychological and psychosocial considerations; comorbidities; the role of clinicians; environment and external influences; broader topics incorporating assisted dying; and moral, ethical and legal issues.

Ten of the 85 proposed questions were rated as being important ($\geq 7/10$) by at least 50% of respondents.

Conclusions

Research questions with the highest levels of consensus were predominately concerned with understanding how and why people make end-of-life decisions, and which factors influence those decisions. Dissemination of these findings alongside a focused examination of the existing literature may be the most effective way to add evidence to the ongoing debate around assisted dying.

Word count

Abstract: 289

Main text: 4472

Strengths and weaknesses

- To our knowledge, this is currently the only attempt to identify the main areas of uncertainty and subsequent research priorities to inform the ongoing debate around assisted dying.
- All research questions were generated and refined by a group of interested parties from both sides of the debate, most of whom considered themselves to be very knowledgeable about the topic.
- The research priorities were collectively identified by health and social care professionals, researchers, campaigners, patients and carers, rather than by researchers alone.
- The response rate and overall level of consensus as defined in this study were relatively low.
- Very few of the religious groups that we approached responded to the survey.

Introduction

Public debate around end of life issues has increased in recent years, partly because of demographic changes caused by a rapidly aging population without a corresponding increase in healthy life expectancy.¹ The debate on assisted dying has become particularly prominent, to the extent that individual 'right to die' appeals frequently receive national media coverage²⁻⁴ and the topic has been explored in novels,⁵ movies⁶ and even popular soap opera storylines.⁷

'Assisted dying' is not a legal term, but is typically understood to mean a circumstance in which a chronically or terminally ill person is allowed to end their own life, either by assisted suicide (the patient is given lethal drugs to take themselves) or euthanasia (somebody else administers lethal drugs to the patient).

The majority of jurisdictions that have legalized assisted dying permit assisted suicide (in which the patient must themselves take action to end their own life), though some permit voluntary euthanasia or both. Some form of assisted dying is legal in Columbia, Switzerland, Netherlands, Belgium, Luxembourg, Canada, and the US states of Oregon, Washington, Montana and Vermont.

Assisted dying remains illegal in the UK, with proposed bills for legalization recently having been strongly rejected by both MSPs and MPs in May 2015 and September 2015 respectively.^{8,9}

Contrastingly, legislators in the state of California have agreed to allow doctor-assisted dying from June 2016,¹⁰ while the Canadian government proposed similar legislation in April 2016.¹¹

The debate around any proposed legal changes is strongly polarized, and a number of opposing arguments have been put forward. Many arguments focus on moral, ethical, religious or legal issues and are often strongly informed by pre-existing beliefs. Far fewer arguments are based on objective empirical evidence, or else evidence is used only selectively. As such, debates often end in impasse.

Some key areas of debate include: individual autonomy, patient choice, the roles of doctors and relatives in assisting death, the nature of 'unbearable' illness, the impact of availability and efficacy of palliative care, the effectiveness of safeguards to protect vulnerable people, and the extent of 'unseen' assisted dying/suicide within the current system. Each of these raises questions about "what we know" about the current state of affairs, whether there is a need for further objective

evidence to illuminate the debate on assisted dying, and if so, what focus and form this evidence might take.

We therefore conducted a modified Delphi exercise with experts and interested parties to identify the main areas of uncertainty and subsequent research priorities to inform the ongoing debate around assisted dying.

Method

Design

The Delphi technique is a structured and iterative method for collecting anonymous individual opinions from a panel of topic experts where a consensus is required. The basic principle is for the panel to receive successive questionnaires, each one containing the anonymous responses to the previous round, and for them to modify their responses until a consensus is reached.¹²⁻¹⁴ The method has been used to identify research priorities in a number of different topic areas.¹⁵⁻¹⁷

For the debate around assisted dying, this approach allowed respondents to generate a number of relevant research questions and then identify those of the highest priority. The aim was to identify any commonly agreed areas of perceived uncertainty, regardless of prior stance or beliefs.

In the first of two rounds, participants were invited to suggest areas of uncertainty that could be addressed by research:

“A number of arguments have been made both for and against changing the law around assisted dying. Some of these are moral or ethical arguments, others are legal arguments and others are medical or pragmatic arguments. These different arguments require differing degrees of supporting evidence. The aim of this study is to identify the most important areas where the facts are unknown or uncertain, either because there is no evidence or because the evidence we have is limited.

In this first stage, we would simply like you to suggest - as concisely as possible - where you think there is a need for either new evidence or a better summary of the existing evidence in this area.”

Participants were presented with illustrative examples of research questions and then provided space to suggest their own (see Appendix 1 for a complete list of questions proposed by the respondents). No restrictions were placed on the number of suggestions a participant could make. After the first round was closed, thematic analysis was used to de-duplicate, code, and group items.¹⁸

In the second round, participants were asked to rate the importance of each research question on a scale from 1 (not at all important) to 10 (extremely important). Participants were encouraged to give a low score to questions that have already been fully answered or are of little interest, but to highly score questions where there is still uncertainty and where research is urgently needed. Questions receiving a score of ≥ 7 points on the ten-point scale were interpreted as having high importance. A ‘No opinion’ option was also provided. Since the respondent population in this second round was unlikely to be identical to that in the first round, participants in both stages were briefly asked to rate their current level of knowledge and give their stance on hypothetical changes in the law.

Results of both rounds were shared with respondents.

Questionnaires were administered electronically using on-line survey software Survey Monkey (www.surveymonkey.com).

Participants

Experts and parties interested in the subject of end of life care and/or assisted dying (clinicians, charities, religious groups, specialist research groups, think-tanks, pressure groups, patient and carer representatives) were approached to participate.

An initial list of individuals, organisations and groups was identified through searches of both academic, government and mass media publications, including the 2012 report published by the Commission on Assisted Dying in which a broad range of interested parties submitted evidence.¹⁹ In addition, pro- and anti-assisted dying groups (e.g. Dignity in Dying, Living and Dying Well, Care Not Killing) were contacted directly. These sources identified a range of groups and individuals who have previously shown an interest in the legal status of assisted dying in the UK.

Email addresses were collected from personal contact lists and publicly available sources (e.g., organisational websites). All emails were personalised to individuals. Groups were encouraged to disseminate the invitation to their members via their own websites, electronic mailing lists and newsletters. Anyone responding to a link cascaded by an original contact was added to the contact list and sent a questionnaire. All contacts were assured confidentiality, with the aim of encouraging participation and openness.

All contacts were invited to both rounds of the survey, including first-round non-responders (unless they had chosen the option to withdraw from further contact).

In order to assess representation from different stakeholder groups and identify any differences in the responses between them, participants were also asked to categorise and/or briefly describe their background, rate their self-perceived level of knowledge on the topic of assisted dying, and give their general prior opinion on the issues of assisted dying and voluntary euthanasia.

Backgrounds were categorized as: clinician, patient, carer/ex-carer, patient representative, researcher, campaigner, legislator, or other. Respondents could choose one or more categories and were encouraged to provide further details where necessary.

Respondents' prior opinions were elicited with three questions on hypothetical changes to the existing law. These asked: "Do you think that under certain defined circumstances, the law should allow..."

1. ...assisted suicide (providing someone with the means to end his or her own life)?
2. ...voluntary euthanasia (ending another person's life at their own request) by a doctor?
3. ...voluntary euthanasia (ending another person's life at their own request) by a close family member?

These three scenarios reflect proposed or existing legal arrangements in jurisdictions outside the UK and reflect the chief options contested in the debate around assisted dying. Respondents could reply "Yes", "No", "Don't know", or "Rather not say".

Questionnaires

Questionnaires were piloted before distribution, resulting in minor wording changes to improve clarity. Round one was initially 'open' for responses for two weeks, though this was extended by a week after several contacts requested extra time to formulate their questions. Round two was open for three weeks. Reminder emails were sent to all contacts approximately one week before the close of each round. A mixture of pre-specified and free text responses were used to facilitate ease of response and analysis of data. In order to ensure that sufficient data were collected and to prevent inadvertent missing data, 'pick list' questions were made mandatory. It was not mandatory to complete the free text boxes.

Analysis

All responses were collected in 'Survey Monkey' for initial tabulation and analysis. Subsequent analyses and outputs were produced in Excel. Descriptive statistics were calculated and used to investigate the distribution of scores. An initial consensus level was set at a score of ≥ 7 points on the ten-point importance scale from $\geq 50\%$ of respondents. Where a respondent did not provide a score, this value was recorded as missing; there was no imputation of missing values.

The relationship between respondents' prior position and scoring of higher consensus questions was explored. Variances were unequal across groups, with small numbers of observations for some questions, precluding the calculation of meaningful hypothesis tests. Therefore these data were displayed graphically.

Ethical approval

Invitees were promised anonymity and submission of completed questionnaires was taken as implied consent. The research was approved by the University of York Health Sciences Research Governance Committee.²⁰

Results

A total of 110 individuals and organizations were initially invited to participate. An additional 16 contacts were suggested by first round respondents, and a further 31 were suggested by second round respondents, all of whom were also invited to participate.

First round

Respondent characteristics

30 (24%) invitees (28 of whom completed all questions) responded to the initial questionnaire that required the formulation of research questions. Seven invitees declined the invitation, one email was undeliverable, and the remainder provided no response.

75% of respondents rated themselves as being "very knowledgeable" or "expert" on the topic of assisted dying (Table 1). Only one respondent considered themselves to have "limited knowledge".

Respondent backgrounds included health and social care professionals (palliative care, oncology, nursing, general practice, public health, social work), researchers, campaigners, and patients or their carers/representatives (Table 2).

A large majority of respondents (93%) were UK-based, though responses were also received from elsewhere in Europe, including Belgium and Switzerland.

Opinions on assisted dying and voluntary euthanasia were mixed. In response to the question “Do you think that under certain defined circumstances, the law should allow assisted suicide (providing someone with the means to end his or her own life)?”, 50% of respondents answered “Yes”, 36% answered “No” and 14% responded that they did not know or would rather not say. A similar pattern of responses was seen for voluntary euthanasia by a doctor (Yes 46%, No 40%, Don’t know/Rather not say 14%), though a majority of respondents were opposed to voluntary euthanasia by a close family member (60% vs. 21% in favour and 18% uncertain or unwilling to respond).

Table 1: Respondents' self-rated knowledge

How would you rate your own knowledge on the topic of assisted dying?	Round 1		Round 2	
	Count	%	Count	%
Know nothing	0	0	1	2.9
Limited knowledge	1	3.6	2	5.7
Reasonably knowledgeable	6	21.4	14	40.0
Very knowledgeable	15	53.6	12	34.3
Expert	6	21.4	6	17.1
TOTAL	28	100	35	100

Table 2: Respondents' background

How would you describe your background?	Round 1	Round 2
Clinician	11	9
Patient	2	2
Carer or ex-carer	1	9
Patient representative	5	4
Researcher	8	9
Campaigner	5	9
Legislator	0	0
Other	5	5
Additional background details (where reported)		
Palliative care	4	5
Hospital consultant	4	1
Oncology	1	1
Public health	1	0
Gerontology	0	1
Nursing	1	0
General practice	1	2
Psychiatry	0	1
Right to die group/organisation	3	2
Policy advice	2	0
Social work	1	1
Charity sector	2	1

Law	0	1
Chaplaincy	0	1
Veterinary medicine	0	1
Relative	0	1

Respondent-generated research questions and themes

Respondents made 102 separate suggestions for research questions. Thematic analysis identified thirteen interrelated themes (Table 3)

Table 3: Research question themes

Theme	Number of suggested questions
Palliative care/symptom control	7
Patient characteristics, experiences and decisions	13
Families and carers	3
Society and the general public	7
Arguments for and against assisted dying	4
International experiences / analysis of existing national data	18
Suicide	2
Mental health, psychological and psychosocial considerations	9
Comorbidities	3
The role of clinicians	7
Environment and external influences	4
Broader topics incorporating assisted dying	2
Moral, ethical and legal issues	6

Where two or more responses expressed an identical research question in different ways, the most concise formulation was kept. Where two or more responses expressed similar but not necessarily identical questions, both versions were retained for the second round. Some responses were categorized as “Non research questions” where they posed largely moral/ethical questions or statements of opinion that could not be rephrased without the risk of changing the meaning intended by the respondent (e.g. “Moral arguments around autonomy and dignity”).

After de-duplication, a total of 85 questions in the 13 categories were sent to participants to be rated in the second round (see Appendix 1).

Second round

Respondent characteristics

A greater number of participants (n=39; 26%) responded in the second round than the first. Respondent backgrounds, where reported, were similar between rounds, though a greater number of respondents in the second round categorized their backgrounds as “carer / ex-carer”, “campaigner” or both (Table 2).

Participants were again asked to rate their current level of knowledge and give their stance on hypothetical changes in the law. Compared with the first round, a smaller proportion of respondents (51.4%) rated themselves as being “very knowledgeable” or “expert” on the topic of assisted dying than did in the first round (Table 1).

A greater proportion of respondents were sympathetic to assisted dying and voluntary euthanasia by a doctor than in the first round (74% vs 50% and 66% vs 46% respectively); the majority remained opposed to voluntary euthanasia by a close family member (54%; Figure 1).

Respondent ratings of research questions

A median of 32 ratings (range 23 to 36) were available for each of the 85 questions.

The distribution of scores was negatively skewed (i.e. in favour of high ratings of importance) for all questions, but varied widely between individual respondents (76 questions received ratings ranging from 1 to 10 points). Appendix 1 shows box-and-whisker plots summarizing the mean, median, range and interquartile range values for each question.

The level of consensus among respondents on whether a question was important (i.e. score ≥ 7) ranged from 4% to 56% (see appendix 2 for details)

Summary of highest priority questions

Ten of the 85 rated questions met the consensus level of receiving a score of ≥ 7 points from $\geq 50\%$ of respondents. These are shown in Table 4.

Table 4: Questions rated $\geq 7/10$ by at least 50% of respondents

Primary theme	Proposed research question	Consensus
Families and carers	What are the effects of carer burden on requests for assisted dying?	55.9%
Arguments for and against assisted dying	Understanding better why some patient groups are strongly opposed to assisted suicide - what are their concerns, could these be mitigated?	54.3%
Comorbidities	Given the progression of dementia, when should end of life care be discussed with the person with dementia and who should initiate this discussion?	53.3%
Mental health, psychological and psychosocial issues	What are the triggers for requesting assisted dying?	52.9%
Personal characteristics, experiences and decisions	How do the views of people considering euthanasia/physician assisted suicide with a spectrum of conditions develop over time - especially those not near the end of life?	52.8%
International experiences / analysis of existing data	Does international experience confirm or lay to rest concerns that vulnerable individuals will be pressurized to avail themselves of assisted dying?	51.5%
Personal characteristics, experiences and decisions	Why do people consider going to Dignitas - is it fear of dying, pain, control?	50%
Personal characteristics, experiences and decisions	What would enhance a person's quality of life after diagnosis of a terminal illness, how do they define 'quality of life' and what are factors they take into consideration in assessing it?	50%
Broader topics	How to operationalise concepts such as "unbearable suffering"?	50%

Moral, ethical and legal issues	The best alternative care pathways for 'end of life' (rather than depriving the patient food and drink and allowing them to starve)?	50%
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The question with the single highest level of consensus was:

“What are the effects of carer burden on requests for assisted dying?” (55.9%)

This was one of several highest-level consensus questions specifically concerned with understanding how and why people make end-of-life decisions, and which factors influence those decisions:

“How do the views of people considering euthanasia/physician assisted suicide with a spectrum of conditions develop over time - especially those not near the end of life?” (52.8%)

“What are the triggers for requesting assisted dying?” (52.9%)

“Why do people consider going to Dignitas - is it fear of dying, pain, control?” (50%)

What would enhance a person’s quality of life after diagnosis of a terminal illness, how do they define 'quality of life' and what are factors they take into consideration in assessing it” (50%)

These questions imply a need for robust qualitative evidence about individuals’ motivations, experiences and decisions, while another question identified the analysis of existing evidence from jurisdictions in which some form of assisted dying is already legal as an area of importance:

“Does international experience confirm or lay to rest concerns that vulnerable individuals will be pressurized to avail themselves of assisted dying?” (51.5%)

One question raised a general methodological issue for end-of-life researchers:

How to operationalise concepts such as "unbearable suffering"?” (50%)

Two consensus items touched on issues broader than simply obtaining good research evidence, raising questions about optimal planning and decision-making at the end of life:

“Given the progression of dementia, when end of life care should be discussed with the person with dementia and who should initiate this discussion?” (53.3%)

“What are the best alternative care pathways for 'end of life' (rather than depriving the patient food and drink and allowing them to starve)?” (50%)

Finally, one proposed research question was not concerned with understanding people at the end of life, but those who take a particular position on the issue of assisted dying:

“Understanding better why some patient groups are strongly opposed to assisted suicide - what are their concerns, could these be mitigated?” (54.3%)

Relationship between respondents' views on assisted dying and their perceived importance of research questions

Figures 2, 3, and 4 illustrate the relationship between respondent views on various forms of assisted dying and mean ratings for questions with $\geq 50\%$ consensus. The pattern of scores in Figure 2 suggests that respondents with no fixed position on assisted suicide (i.e. "Don't know") consistently gave higher ratings to the research questions than did respondents with a fixed position (i.e. "Yes" or "No"). As might be expected, respondents with less certainty tended to value research evidence more highly.

The pattern of ratings was not consistent for all views on assisted dying (assisted suicide, doctor-assisted voluntary euthanasia, family-assisted euthanasia), though mean ratings tended to be slightly lower among respondents who were against any hypothetical changes in the law (Figures 2, 3 and 4).

Discussion

To our knowledge, this is currently the only attempt to identify the main areas of uncertainty and subsequent research priorities to inform the ongoing debate around assisted dying. All research questions were generated and refined by a group of interested parties from both sides of the debate, most of whom considered themselves to be very knowledgeable about the topic.

New research ideas are frequently generated by research professionals who have an interest in a topic area. While researchers may be able to identify gaps in the evidence, they may not be best placed to determine which areas are most urgently in need of further research. Approaches such as the James Lind Priority Setting Partnerships aim to tackle treatment uncertainties by consulting a wider range of participants in order to identify research priorities. Such partnerships consist of at least one patient/carer organisation and at least one clinician organisation.²¹

This Delphi consultation incorporated a diverse set of experiences, including those of health and social care professionals, researchers, campaigners, patients and carers from a predominately UK setting. Consequently, the research priorities identified here may more closely reflect those of value to wider society. For example, researchers might be interested in analyzing data collected in jurisdictions where assisted dying is legal. However, while research questions about cross-country comparisons were most commonly suggested by panel members, only one of these emerged among the highest-priority questions based on consensus score.

The results raised a number of important questions about end-of-life issues that were broader than just the topic of assisted dying. For example, clear definition and measurement of concepts such as 'quality of life' and 'unbearable suffering' in this setting are fundamental to understanding end-of-life issues, yet the panel of experts considered the evidence to be lacking in this area. The NIHR 'Palliative and end of life care Priority Setting Partnership' (PeolcPSP) has looked at end of life issues more broadly and identified a set of unanswered questions around provision and access to palliative care and the benefits of Advance Care Planning that complement those identified in the current exercise.²² Some issues - such as concerns about how to listen to and incorporate patient preferences - overlap with the priorities identified here. Interestingly, the PeolcPSP also received a number of comments and questions outside its intended scope.

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3 The overall level of consensus as defined in this study was relatively low (4% to 56%). However, wide
4 variation in second round scores between items suggested that respondents were able to distinguish
5 pertinent research questions from untestable hypotheses and statements of opinion. Therefore it
6 seems that most respondents understood the aim of the project, and were focused on identifying
7 areas of uncertainty that would benefit from empirical investigation. Further, it seems that the
8 consensus threshold applied here (at least half the respondents giving ≥ 7 points) was able to identify
9 the highest priority questions.
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12 The response rate to the Delphi (around 25%) was relatively low, but compares favorably to other
13 surveys that have recruited doctors.^{23, 24} A low response might have been expected, given the onus
14 on respondents to formulate their own research questions; the level of time and effort required for
15 this may have been a barrier for participants who might otherwise have responded to a simple 'tick-
16 box' questionnaire. However, a fundamental objective of this process was to obtain research
17 questions from experts and other interested parties, rather than have them imposed by researchers.
18 Similar future surveys should consider engaging participants as early in the process as possible, and
19 attempt to sustain participant enthusiasm to overcome such barriers. Alternatively, questions might
20 be initially generated through interviews or focus groups.
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24 Over 90% of respondents were based in the UK, with the remainder from elsewhere in Europe.
25 There were no respondents from other regions, in particular the US states where assisted dying
26 legislation has been enacted. This would raise concerns about possible unrepresentativeness if the
27 identified high-consensus questions related specifically to medico-legal issues. However, the highest-
28 consensus questions identified here relate to how and why people make end-of-life decisions, which
29 are more likely to be of universal concern. Nevertheless, we acknowledge that the relative emphasis
30 placed on such questions may partly depend on cultural context.
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34 Whereas large sample sizes are important for questions of precision, the aim of the Delphi exercise
35 was to identify consensus among a diverse group of interested individuals. In this case, obtaining an
36 appropriate sampling frame is perhaps more important. As well as including participants from a
37 variety of professions, we approached groups and individuals with opposing opinions on the subject
38 of assisted dying. While respondents from both sides of the debate contributed to both stages of the
39 survey, there was a slight predominance of respondents in favour of assisted dying. Very few of the
40 religious groups that we approached responded to the survey, so this may have had an influence on
41 the ratings (i.e. through underrepresentation of anti-assisted dying opinion), and might partly
42 explain high importance ratings for the question "Understanding better why some patient groups
43 are strongly opposed to assisted suicide - what are their concerns, could these be mitigated?".
44 However, the other highly-rated questions do not appear to have an overtly pro- or anti-assisted
45 dying stance, but rather address important areas of uncertainty. The distribution of scores did not
46 show a clear influence of prior stance on question ratings: a slight tendency for lower scores among
47 those respondents against a change in the law was observed, though for most questions the scores
48 did not differ greatly. It is plausible that people who consider the current legal position to be
49 adequate are less likely to believe there is a need for research than people who are unsure or favour
50 a change in the law.
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Recommendations for future research

The logical next step would be to address the priority questions identified from this process. This should be done in the first instance by examining the existing evidence to further refine the design of any future research. There is some available evidence relevant to some of the questions identified here (for example, on views of patients and carers),²⁵ but a number of the questions have not been addressed directly or systematically. Before undertaking any new primary research, one or more systematic reviews of the existing evidence focusing on the themes and questions identified here may be worthwhile. For example, a review of qualitative evidence specifically concerned with the influence of dementia on patient and carer views related to assisted dying would be of value. Though lower priority, this may be supplemented by a systematic review of the international evidence to determine the fate of vulnerable people in jurisdictions with legalized forms of assisted dying, which remains an area of major contention. A well conducted systematic review could provide an impartial and comprehensive overview of the evidence, making explicit its relative strengths and weaknesses in relation to the well-worn arguments in this area. If uncertainties still remain, the review could make clear and precise recommendations about where new primary research is needed.

Conclusion

This consultation revealed a number of important uncertainties around the debate on assisted dying and end-of-life issues more broadly. Eighty-five unique research questions were suggested by a broad range of interested parties with high levels of topic expertise. Research questions with the highest levels of consensus were predominately concerned with understanding how and why people make end-of-life decisions, and which factors influence those decisions. Dissemination of these findings alongside a focused examination of the existing literature may be the most effective way to bring objective research evidence into the ongoing debate around assisted dying.

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Contributorship statement

Mark Rodgers contributed to the design of the study, collected, analysed and interpreted the data, drafted, revised and approved the manuscript.

Alison Booth contributed to the design of the study, interpreted the data, and revised and approved the manuscript.

Gill Norman contributed to the design of the study, interpreted the data, and revised and approved the manuscript.

Amanda Sowden contributed to the design of the study, interpreted the data, and revised and approved the manuscript.

Competing interests

The authors have no competing interests to declare

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Data sharing statement

No additional data available

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References

1. Cracknell R. *The ageing population: Key issues for the 2010 Parliament*: House of Commons Library Research; 2010. Available from: <http://www.parliament.uk/business/publications/research/key-issues-for-the-new-parliament/value-for-money-in-public-services/the-ageing-population/>
2. BBC News. Husband speaks out on wife's assisted suicide. [serial online] 2012. [cited Available from: <http://www.bbc.co.uk/news/uk-england-suffolk-17550982>
3. BBC News. *Right-to-die man Tony Nicklinson dead after refusing food*. 2012. [cited Available from: <http://www.bbc.co.uk/news/uk-england-19341722>.
4. ITV News. *Hope remains despite Supreme Court rejection of right to die campaign*. 2014. [cited Available from: <http://www.itv.com/news/calendar/2014-06-25/hope-remains-despite-supreme-court-rejection-of-right-to-die-campaign/>.
5. Moyes J. *Me Before You*. London: Penguin Books; 2012.
6. Moore, JD. (Director), Eisen, A. (Writer). *Youth in Oregon*. Sundial Pictures; 2016.
7. ITV News. *Coronation Street shows controversial right to die scene*. 2014. [cited Available from: <http://www.itv.com/news/2014-01-20/coronation-street-controversial-assisted-suicide-scene-hayley-cropper/>.
8. *Assisted Suicide (Scotland) Bill*. The Scottish Parliament; [cited 2016 7th April]. Available from: <http://www.scottish.parliament.uk/parliamentarybusiness/Bills/69604.aspx>.
9. *Assisted Dying (No.2) Bill 2015-16*. UK Parliament; [cited 2016 7th April]. Available from: <http://services.parliament.uk/bills/2015-16/assisteddyingno2.html>.
10. Clodfelter R, Adashi E. The Liberty to Die: California Enacts Physician Aid-in-Dying Law. *JAMA: Journal of the American Medical Association* 2016;315:251-2. Available from: <http://jama.jamanetwork.com/article.aspx?articleid=2482335>
11. *Government introduces legislation to provide medical assistance in dying*. Government of Canada; [cited 2016 29th April]. Available from: <http://news.gc.ca/web/article-en.do?nid=1051809&tp=1>.
12. Hsu C, Sandford B. The Delphi Technique: Making Sense of Consensus. *Practical Assessment Research & Evaluation* 2007;12:1-8.
13. Murphy E, Black N, Lamping D, McKee C, Sanderson C. Consensus development methods, and their use in clinical guideline development: a review. *Health Technology Assessment* 1998;2:1-88.
14. Sinha I, Smyth R, Williamson P. Using the delphi technique to determine which outcomes to measure in clinical trials: recommendations for the future based on a systematic review of existing studies. *PLoS Medicine* 2011;8:e1000393.
15. Cresswell K, Panesar S, Salvilla S, Carson-Stevens A, Larizgoitia I. Global Research Priorities to Better Understand the Burden of Iatrogenic Harm in Primary Care: An International Delphi Exercise. *PLoS Medicine* 2013;10:e1001554.
16. Rankin G, Rushton A, Olver P, Moore A. Chartered Society of Physiotherapy's identification of national research priorities for physiotherapy using a modified Delphi technique. *Physiotherapy* 2012;98:260-72.
17. Wielenga J, Tume L, Latour J, van den Hoogen A. European neonatal intensive care nursing research priorities: an e-Delphi study. *Archives of Disease in Childhood-Fetal and Neonatal Edition* 2015;100:F66-71.
18. Guest G, MacQueen K, Namey E. *Applied thematic analysis*. Thousand Oaks: SAGE; 2011.
19. The Commission on Assisted Dying. 2011. [cited Available from: http://www.demos.co.uk/files/476_CoAD_FinalReport_158x240_I_web_single-NEW_.pdf?1328113363.
20. University of York Health Sciences Research Governance Committee. 2015. [cited Available from: <http://www.york.ac.uk/healthsciences/research-information/rsg/>.
21. James Lind Alliance. *Priority Setting Partnerships*. 2015. [cited Available from: <http://www.lindalliance.org/Priority%20Setting%20Members.asp>.

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3 22. Palliative and end of life care Priority Setting Partnership. *Putting patients, carers and clinicians*
4 *at the heart of palliative and end of life care research*. 2015. [cited Available from:
5 http://www.palliativecarepsp.org.uk/wp-content/uploads/2015/01/PeolcPSP_Final_Report.pdf.
6
7 23. Baruch Y. Response rate in academic studies-A comparative analysis. *Human Relations*
8 1999;52:421-38.
9 24. Scott A, Jeon S, Joyce C, Humphreys J, Kalb G, Witt J. A randomised trial and economic evaluation
10 of the effect of response mode on response rate, response bias, and item non-response in a survey
11 of doctors. *BMC Medical Research Methodology* 2011;11:126.
12 25. Hendry M, Pasterfield D, Lewis R, Carter B, Hodgson D, Wilkinson C, et al. Why do we want the
13 right to die? A systematic review of the international literature on the views of patients, carers and
14 the public on assisted dying. *Palliative medicine* 2013;27:13-26.
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Appendices

Appendix 1: Box and whisker plots of all responses to questions rated in round 2

Appendix 2: Percentage consensus on importance of questions listed in round 2

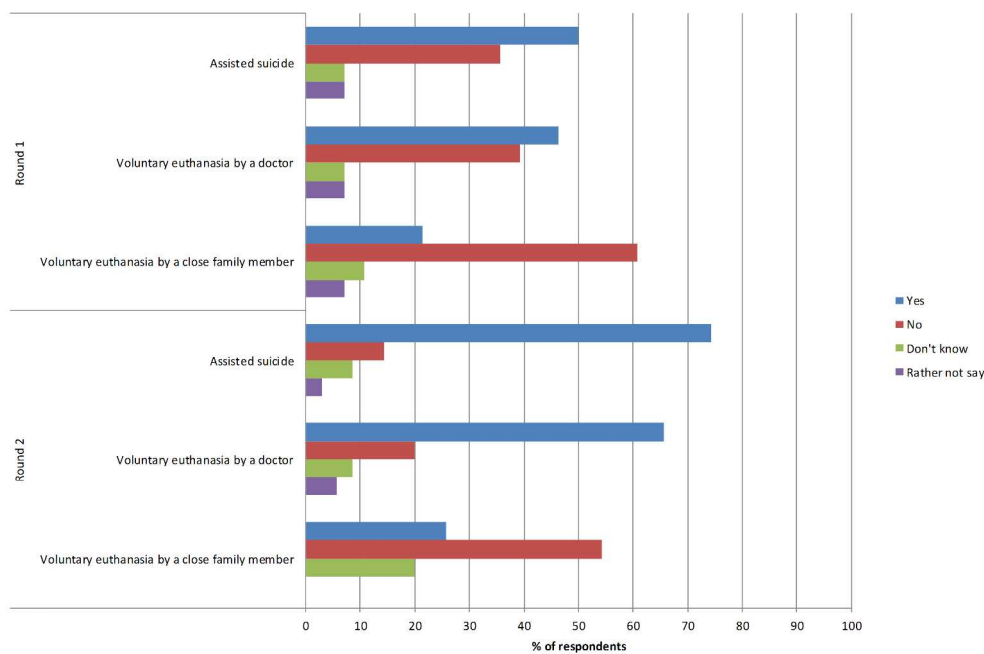


Figure 1: Respondent support for a change in the law
269x182mm (300 x 300 DPI)

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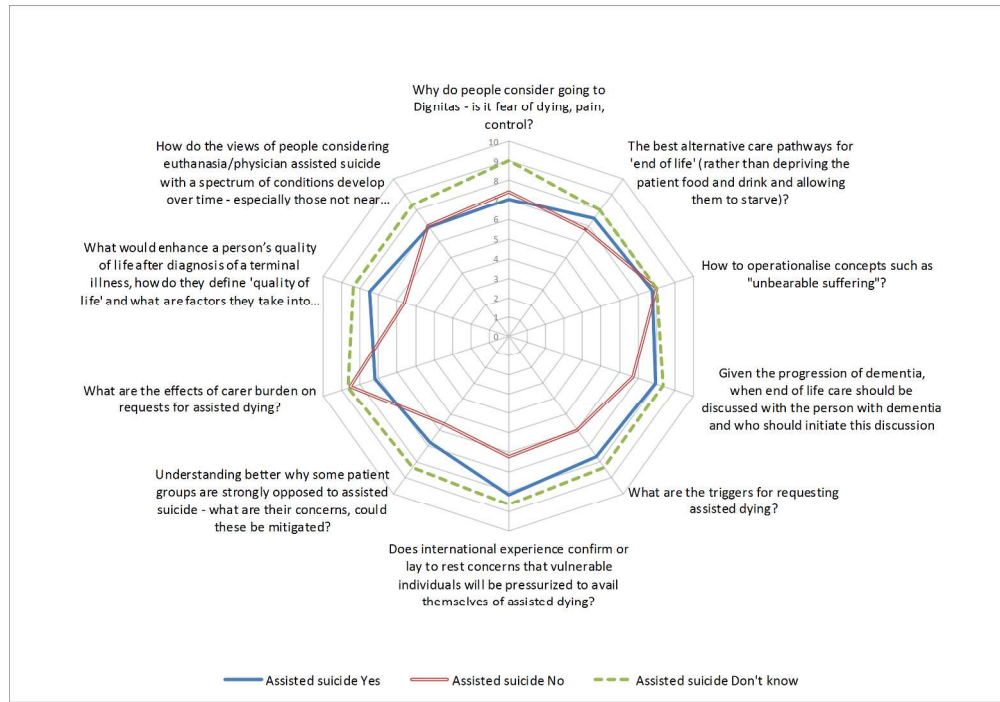


Figure 2: Relationship between respondent views on assisted suicide and mean rating for highest consensus questions
245x170mm (300 x 300 DPI)

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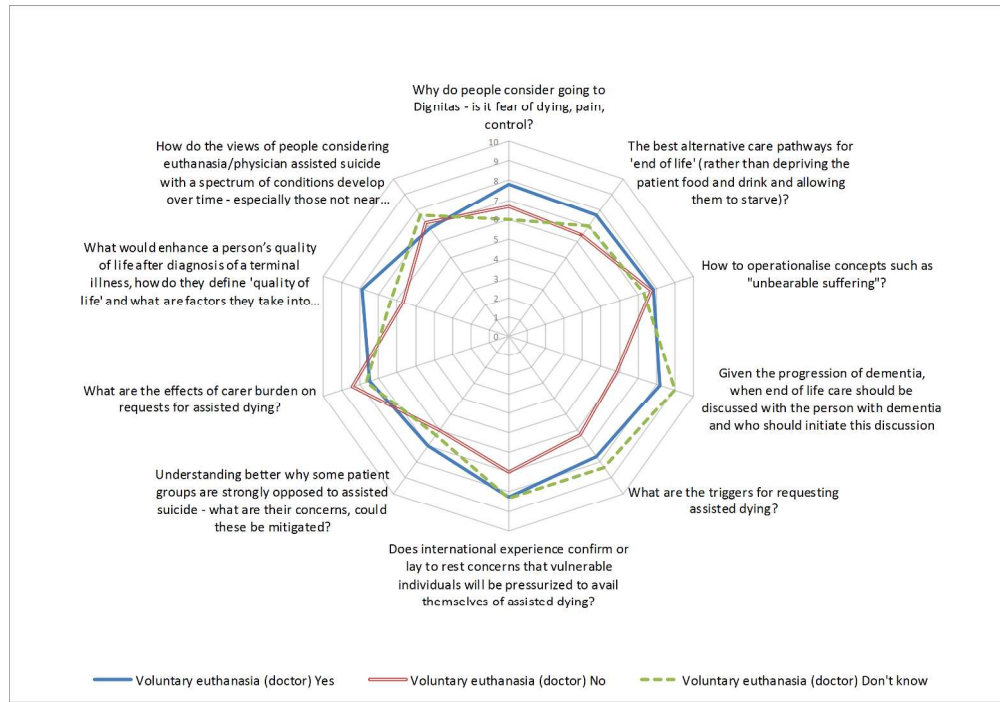


Figure 3: Relationship between respondent views on doctor-assisted voluntary euthanasia and mean rating for highest consensus questions
245x170mm (300 x 300 DPI)

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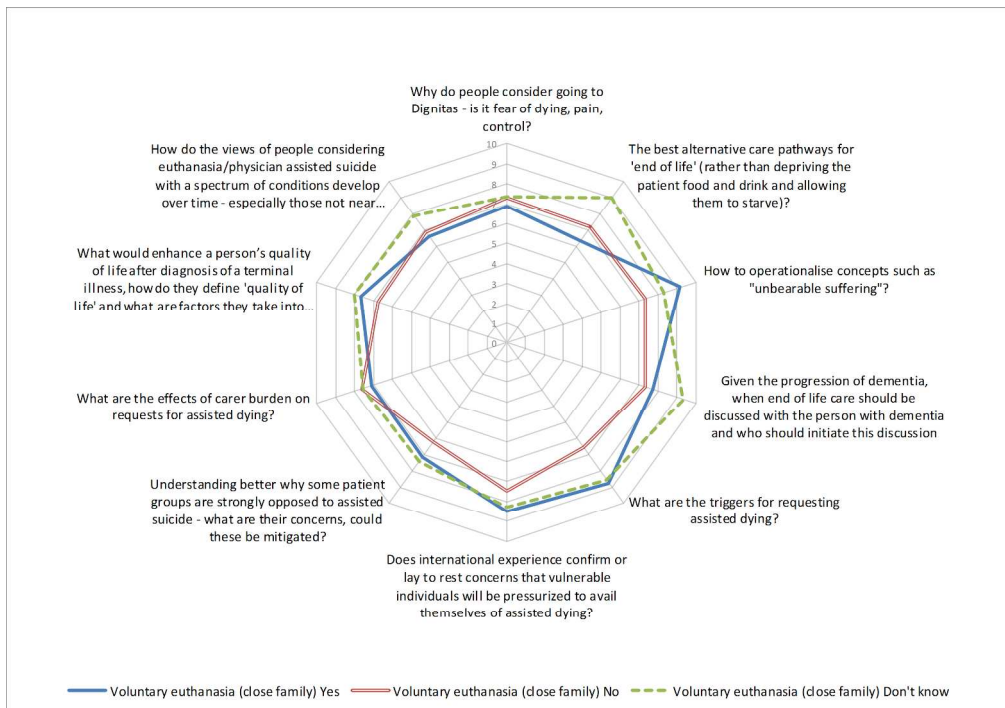


Figure 4: Relationship between respondent views on family-assisted voluntary euthanasia and mean rating for highest consensus questions
245x172mm (300 x 300 DPI)

Review only

Appendix 1: Box and whisker plots of all responses to questions rated in round 2

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Appendix 2: Percentage consensus on importance of questions listed in round 2

