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1 Close person spill-overs in end of life care: using hierarchical mapping to identify 2 whose outcomes to include in economic evaluation

3 Abstract

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11 Background

12 Guidelines for economic evaluation often request that costs and outcomes beyond the patient are
13 captured; this can include carers and also other affected parties. End of life care is one context
14 where impacts of care spill over onto those other than patients, but there is little evidence about
15 who should be included within economic evaluation. The purpose of this paper is to examine (i) how
16 many people are close to those at end of life (ii) their characteristics and (iii) what influences
17 network size at end of life.

18 Methods

19 In-depth interviews were conducted with 23 participants who were either recently bereaved or had
20 somebody close to them currently receiving end of life care. Interviews were used in conjunction
21 with hierarchical mapping to explore the network size, composition and influences upon these
22 networks. Interviews were transcribed verbatim. Descriptive statistics were used to analyse the
23 hierarchical maps and this information was combined with a constant comparative analysis of the
24 qualitative data.

25 Results

26 On average, close-person networks at end of life contained eight individuals, three of whom were
27 rated as being 'closest'. These were typically family members although in a small number of cases
28 non-family members were included amongst the closest individuals. There was variation in terms of
29 network composition. Qualitative analyses revealed two key influences on network size: death
30 trajectory (those with cognitive problems/diseases towards end of life had smaller networks) and
31 family size (larger families had larger networks).

32 Conclusion

33 The findings of this paper have important implications for researchers wishing to include those
34 affected by end of life care in economic evaluation. Focussing on the three closest individuals would
35 be a key starting point for economists seeking to capture spill-overs whilst a truly societal

1 perspective would require looking beyond proximal family members. This paper further discusses
2 the implications of including close-persons in economic evaluation for decision makers.

4 Key points:

- 5 - We use hierarchical mapping alongside in-depth interviews to examine who and how many
6 are close to those at end of life and could be considered for inclusion in economic
7 evaluation.
- 8 - On average, networks at end of life contained eight people, of which three were rated as the
9 closest. Family members were typically closest, although this was not always the case.
10 Likewise, geographical proximity was not necessary to maintain close person relationships.
- 11 - Qualitative analyses revealed two key influences on network size: death trajectory (those
12 with cognitive problems/diseases towards end of life had smaller networks) and family size
13 (larger families had larger networks). These findings have important implications for
14 researchers wishing to include close-persons affected by end of life care in economic
15 evaluation.

16 1. Background

17 Economic evaluation involves comparison of the costs and consequences of two or more alternative
18 courses of action [1]. Historically in the UK, the National Institute for Health and Care Excellence
19 (NICE) has recommended that the primary focus of economic evaluation should be on outcomes for
20 the patient [2–4] and carer when relevant, alongside costs from a health and personal social services
21 perspective [3]. Other countries such the Netherlands have broadened their recommended
22 perspective to that of society, whilst the US Panel on Cost-Effectiveness request two reference
23 cases: a health care and a societal perspective [5,6]. For a true societal perspective, all costs and
24 benefits, irrespective of who bears them should be included within the economic evaluation.

25 Within the health economics literature, there have been arguments for including the impacts of
26 people other than the patient within economic evaluation [7–13], particularly when the impact on
27 people other than the patient is significant. Elsewhere, wider impacts extending beyond the patient
28 as a single (and isolated) individual have been described as being ‘too important to ignore’ [8]; a
29 range of measures exist to try to capture spill-over impacts [7,9,14].

30 End of life care (EoLC) has been highlighted as one area where impacts on those close to the dying
31 (‘close persons’) may be particularly relevant [15], as interventions often extend in scope to these
32 close persons [16], and benefits (or harms) may be received by them [17–21]. Despite movement in
33 some countries towards including close persons within economic evaluation, there is little guidance
34 on who should be included and how to identify these people. For example, when discussing health
35 gain, the US Panel on Cost-Effectiveness refer to *‘other affected parties such as caregivers’* [6], whilst
36 the Dutch guidelines state that *‘all relevant costs and benefits, irrespective of who bears the costs or
37 to who the benefits go’* should be included [5]. These issues are not straightforward, and the
38 common approach of restricting impacts to an identified immediate carer or the next of kin may be
39 insufficient to capture relevant impacts.

40 In adopting a broader or societal perspective it is important to know who, and how many people, are
41 close to those at the end of life and therefore who should be assigned the status of ‘close person’ for
42 inclusion within economic evaluation. There is extensive research on social networks more generally
43 and tools exist to measure the strength of relationships [22–28], however there is a dearth of
44 evidence within the end of life context or with a focus on relevance for economic evaluation.

1 Research focusing on older adults more generally has found variable results. A US survey examined
2 network size in community-dwelling adults (n = 2005) aged between 57 and 85 years by classifying
3 networks into: i) core confidantes, ii) those who are important/close, iii) any remaining household
4 members who were not confidantes or very close; on average there were approximately 3.5
5 individuals named as a confidante [29]. Likewise, using hierarchical mapping techniques, Antonucci
6 [30] found that, on average in the United States, those aged over 50 years had 3.5 individuals within
7 their 'inner circle', that is the closest individuals. On average 8.9 individuals were included across
8 their whole close-person network. In contrast, a survey of older people across Europe found
9 networks to be much smaller, with just 2.5 people being included on average within their social
10 network [31]. Networks are not static through life, however, and it is unknown whether social
11 networks change within the end of life context, when co-morbidities and disease progression could
12 conceivably influence social networks. The goal of this paper is to examine who and how many
13 people are close to those at end of life, and what shapes these networks, as a precursor to
14 developing methods to better capture these impacts for economic evaluation.

15 2. Methods

16 In-depth interviews in combination with hierarchical mapping were used to explore networks at the
17 end of life. The study was conducted alongside a project aiming to develop an outcome measure for
18 use with those close to people at the end of life [9], and as part of a broader exploration of the
19 economic evaluation of end of life care more generally [15]. Both University (ERN_12-1338) and
20 NHS Ethical (13/WA/0333) approvals were obtained.

21 2.1 Sampling

22 Recruitment was driven by the needs of the wider project. The participants of interest within both
23 studies were those who were either recently bereaved or had somebody close to them currently
24 receiving end of life care. Close persons were chosen rather than decedents for two primary reasons:
25 (i) not all people at the end of life would be able to participate, for example, those suffering from
26 cognitive impairment, extreme fatigue, or having experienced sudden death; and (ii) by including
27 bereaved people it was possible to account for the whole end of life period.

28 Sampling aimed to capture people who had experienced a variety of trajectories towards death and
29 focused on those already bereaved. Given this, sampling through a single care provider as a main
30 source was not considered appropriate (although this option was pursued as a supplementary
31 source). In practice, the most feasible means of recruiting from a general population was to recruit,
32 in the first instance through the University of Birmingham staff and students, and then to employ
33 snowball sampling. Recruitment through the university was achieved through internal
34 communications, newsletters and posters (see Appendix 1 for an example advert). A second source
35 of recruitment was a single UK adult hospice. In the UK, the aim of hospice care is to improve the
36 lives of people who have an incurable illness from the point of diagnosis until the end of their life
37 [32]. This hospice comprised an inpatient unit and day hospice and was chosen to ensure people
38 currently being cared for at the end of life were included within the study. Prospective participants
39 were identified and recruited at the hospice by a research nurse. Snowball sampling [31] involved
40 asking participants to pass on an information sheet to all those who they thought might be
41 interested in participating. Snowball sampling [32] also had the potential advantage of enabling
42 exploration of how the perceptions of networks varied within clusters. Overall, sampling using this
43 combination of approaches, was intended to provide access to a large and diverse population in
44 terms of age, profession and the lack of any single specific death trajectory.

1 A window of six to 24 months post-bereavement was used for recruitment purposes to satisfy
2 ethical requirements: individuals who were bereaved within six months were excluded from the
3 study [33] as were those who had been bereaved over two years to minimise recall bias. Information
4 sheets were provided to informants before obtaining informed consent. Recruitment continued until
5 it was felt that saturation had been reached for the wider project goal.

6 2.2 Data collection

7 A qualitative approach, combining semi-structured in-depth interviews with hierarchical mapping,
8 was chosen to facilitate the understanding of networks at end of life. The interviews were conducted
9 by AC (Male/MSc./Doctoral Researcher) who had received prior interviewer training and had
10 experience of conducting qualitative research with vulnerable groups. The interviews were
11 conducted at a location of the participant's choice and were digitally audio recorded and then
12 transcribed verbatim and anonymised. Field notes were taken to inform subsequent interviews.

13 To examine the scope of close-person networks, and the influences of end of life on these networks,
14 a hierarchical mapping [27] task was included. Pictorial tools have been found to aid rapport [34]
15 and elicit complex information in a simple manner [36]. A number of pictorial methods such as the
16 Pictor method [37] were considered. Hierarchical mapping was chosen as it best met the needs of
17 this task in terms of its speed, simplicity and intuitive appeal. Hierarchical maps have been
18 successfully used to explore network composition within a range of settings [38–41]. The
19 hierarchical mapping task involves presenting the interviewee with a large piece of paper with
20 concentric circles going out from the centre (see [42] for a full exposition and Appendix 2 for
21 example)). It was explained to the interviewee that the decedent is in the centre, and the circles
22 represent levels of closeness, with the inner circle being closest. Arrow shaped post-it notes were
23 then given to the interviewee and they were asked to write the name and relation to the decedent
24 of all those who were close onto post-it notes (one per note where possible). They were then asked
25 to place the post-it notes onto the hierarchical map, with the tips of the arrow representing the
26 position of the person in terms of closeness to the decedent (see Figure 1 for completed example).
27 Interviewees were given time to reflect and deliberate. The completed hierarchical map provided a
28 visual representation of the close-person network. It was conducted near the beginning of
29 interviews to allow for the probing of close-person networks in the remainder of the interview.
30 Initial questions focused on asking the informant to describe relationships with the decedent and
31 probes then included questions such as: 'has it always been this way?'; 'how did this change in the
32 period towards the end of life?'; 'how did this make you feel'; 'did their illness impact their
33 relationship with other people?'. Once the hierarchical map was completed, it was used to aid
34 further discussion throughout the interview.

35 2.3 Data analysis

36 To examine the size and composition of close-person networks, descriptive statistics were
37 calculated. The identities of those within each hierarchical map were examined, with particular
38 attention paid to those identified as being closest to the decedent. Hierarchical maps were
39 examined iteratively alongside interview transcripts to gain a deep understanding of close-person
40 networks. Constant comparative analysis [41] was conducted in batches of between three and six
41 interview transcripts was conducted. This included developing a coding structure and creating in-
42 depth descriptive accounts [44] to synthesise and systematically analyse each set of interviews.
43 Coding was conducted by the primary researcher (AC) and reviewed by the research team routinely
44 to ensure data were being analysed in a systematic and rigorous way. To examine influences on
45 network size, those with the smallest networks (five or fewer) were compared and hypotheses were
46 generated from these data. This process was repeated with the largest networks (10+). Deviant case

1 analysis [42] was used to examine any 'unusual' hierarchical maps, examining 'outlier' cases in fine
2 detail to highlight possible explanations for differences.

3 Results are presented using quotes to illustrate themes. Quotes indicate the gender of the
4 informant, and the relationship of the decedent to the informant.

5 3. Results

6 The characteristics of the decedents and the interviewees are shown in Table 1. Location of
7 interviews included: participants' own homes, university meeting rooms, and private rooms within
8 the hospice. In total, 24 decedents were included, with a range of conditions and death trajectories,
9 from slow declining to sudden deaths. This includes all five of the main broad disease groups
10 (cancer, disease of the circulatory system, respiratory diseases, mental and behavioural disorders,
11 and disease of the nervous system) [46]. All decedents were adults, with two aged 40-59 years, 10
12 aged 60-79 years, and nine aged 80+ years. Three informants did not specify the age of the
13 decedent. Interviews took between 25 and 80 minutes (average 45 mins), of which a minority (4.5
14 mins) was spent on the hierarchical mapping task.

1 **Table 1: Decedent close-person networks**

Participant ID	Relation of decedent to participant	Decedent's terminal condition	Decedent's age group	Number of people within decedent's network	Relation of Closest person	Number of non-relatives included within network?	Number of people within inner ring of map	Where they placed themselves in the hierarchical map	Time to complete map (mins)
CDX1	Father	Pancreatic Cancer	60-79	7	Spouse	1	5	Inner ring - closest (equal)	4
CDX4	Brother	Lymphoma	40-59	6	Sister	0	4	Inner ring - closest	5
CDX5	Mother	Alzheimer's	60-79	8	Spouse	0	3	Inner ring - second closest	6.5
CDX5	Father	Heart Failure (with COPD)	80+	7	Daughter	0	2	Inner ring - closest	4
CDX6	Friend (female)	Oesophageal Cancer	40-59	20	Multiple - Friends and Family	2	7	Inner ring - closest	4
CDX7	Mother	COPD and Alzheimer's/Dementia	80+	3	Spouse	0	1	Second ring – second closest	2.5
CDX8	Father	Sudden Death - Heart attack	60-79	6	House Keeper	2	2	Second ring – fifth closest	3
CDX10	Father	Death following elective heart surgery complications	Unknown	74	Multiple - all family	0	9	Inner ring – equal closest	7
CDX13	Mother	Motor Neurone Disease	60-79	9	Multiple - Sister, Daughter and 2 Friends	3	9	Inner ring – equal closest	5
CDX14	Mother	TIA/Dementia	80+	4	Brother	0	1	Second ring – second closest	3.5
CDX16	Father	CHD - Death following heart surgery complications	80+	10	Son	3	5	Inner ring - closest	3
CDX17	Grandmother	Pneumonia	80+	8	Daughter	1	3	Inner ring – second closest (equal)	3
CDX18	Mother	Pneumonia	80+	5	Granddaughter	2	3	Inner ring – second closest	4.5
CDX20	Mother	Cancer - Colon/Liver	60-79	12	Daughter	2	3	Inner ring – closest	3.5
CDX21	Father	Undiagnosed - chest complaint	80+	8	Multiple - Daughter and Spouse	1	3	Inner ring – equal closest	3.5
CDX22	Grandmother	Heart Disease	Unknown	10	Daughters	0	4	Inner ring – third closest	5.5
CDX23	Grandmother	Post-fall infections in hospital	80+	10	Spouse	0	2	Second ring – third equal closest	4.5
CDX24	Grandfather	Lymphoma	60-79	7	Multiple - Granddaughter and Spouse	0	5	Inner ring – third equal closest	2.5
CDX25	Father	Cancer - back/spine	60-79	7	Multiple - Daughter and Spouse	0	5	Inner ring – equal closest	7.5
CDX26	Husband	Multiple System Atrophy	60-79	5	Spouse	0	2	Inner ring – equal closest	4.5
CDX27	Mother	COPD	60-79	8	Multiple - Daughter and Carer (Daughter in law)	2	2	Inner ring – equal closest	3.5
CDX28	Grandmother	Parkinson's Disease	80+	8	Spouse	0	2	Second ring – third equal closest	4.5
CDX29	Mother	Sarcoidosis	60-79	15	Multiple - Son and Brother	0	7	Inner ring – equal closest	16.5
CDX31	Mother	Viral Pneumonia + sudden heart attack	Unknown	8	Spouse	1	5	Inner ring – second closest	3

1 **3.1 Network size**

2 Descriptive statistics for the size of the individual decedent networks are shown in Table 2.

3 **Table 2: Network size descriptive statistics**

	Whole close-person map	Number within inner-ring
Median	8	3
Minimum	3	1
Maximum	74	9
Mean	11	3.9
Standard Deviation	13.9	2.3
Skewness	4.4	0.9
Mode	8	2

4

5 There was a wide variety of hierarchical map size. The smallest contained just three individuals while
6 the largest contained 74. Closer examination of this largest network revealed that the decedent was
7 the social hub of the community prior to a sudden death; consequently, 50 friends were listed within
8 the outer circle. This was very much an outlier causing a skewing of the data with the mean
9 substantially higher than the median. The median network size was eight. Across all networks, a
10 median of three individuals were included within the inner circle, that is, the closest individuals.
11 Nine of the 24 decedents had more than one individual listed as closest; in all other cases, just one
12 individual was listed as closest.

13

14 **3.2 Network composition**

15 Perhaps unsurprisingly, family members were prevalent within the close-person networks of those
16 at the end of life and at least one family member featured within every close-person map. Fourteen
17 (60%) networks, however, featured non-family members including friends, a non-family paid carer,
18 and a house-keeper. Five hierarchical maps contained non-family members within the inner circle of
19 the close-person map. Only one (CDX8, see Figure 2) contained no family members within their inner
20 circle. In terms of the person closest to the decedent, where more than one closest person was
21 named, these were different members of the same family in all but one case (a friend, CDX6, listed
22 herself as equal closest). Those listing just one person as closest, listed the decedent's spouse (n=6),
23 the decedent's child (n=5), a sibling (n=2), a grandchild (n=1), and the house-keeper (n=1). The
24 decedent in the case of CDX8 had separated from the family and moved abroad, hence the unusual
25 network composition, with the housekeeper as the closest person.

26

27 The mapping task demonstrates that broader impacts may impact those other than just family
28 members. Although the vast majority of the closest persons were family members, most networks
29 also included individuals who were not relatives. Of note, in two cases, non-relatives were the
30 closest or equal closest individuals. Through the interview process participants expressed a view that
31 impacts were not limited to family members, and that being close can extend beyond immediate
32 family.

1 CDX20 [female, mother]: *I don't think it has to be defined by family or blood, I think people*
2 *who have made great impacts in your life and you've made great impacts in theirs*

3 CDX1 [female, father]: *...I think it's emotional bonds rather than family bonds if that makes*
4 *sense, quite often the two are the same, but not necessarily.*

5 Given this, when considering the evaluative scope, it is important to consider the inclusion of
6 individuals outside the family.

7 3.3 Influences on network size and composition

8 Three main themes arose in relation to network size and composition: the nature of disease and
9 death trajectory; the size of the family; and geographical proximity.

10 The nature of disease and death trajectory of those at end of life appeared to strongly impact upon
11 network size and composition for some people. Three of the four decedents with the smallest
12 networks suffered from dementia conditions that impacted their cognitive abilities. Participants
13 described how the nature of such diseases led to distancing within the close-person networks.

14 CDX5 [female, mother and father]: *...So, even though my mum was my friend growing up*
15 *and then obviously the disease I felt distanced us*

16 CDX7 [female, mother] *...so my relationship with her changed because I pulled back a lot*
17 *because I could not....if I got too close to her, she'd either hurt me, not physically or although*
18 *she might have had a go, I just got tired of being hurt by it.*

19 In contrast, none of the six participants with the largest close-person networks (over 10 close-
20 persons) had deaths that meant they were cognitively compromised until they were very close to
21 death. Three of these had relatively sudden deaths, two died from conditions that did not impact
22 mental state until the final weeks before death and the final large network belonged to somebody
23 still receiving hospice care at the time of the interview who remained fully cognitively aware.

24 A second key feature relating to network size was family size. All six of the largest networks featured
25 many family members, from seven to 15. In contrast, few family members appeared within the
26 smallest networks, with one informant explicitly citing the lack of a large family.

27 CDX18 [female, mother]: *...We are a very relation-short family*

28 Informants also talked about the influence of geographical proximity on network size and
29 composition, although there were differing views. Some informants felt that being far away from a
30 network member may hinder the maintenance of close-person relations towards the end of life.

31 CDX1: *...my personal view of the world is that it's quite difficult to be close to somebody that*
32 *you're a long way away from or relatively long way away, I mean it's all relative*

33 CDX22: *...I'm close to my cousin and she lived in Belgium, and ...I think it does have an impact,*
34 *it's harder to be closer when they're living somewhere else because obviously all my friends*
35 *who live locally, I'm really glad...I see them all the time so we've got a stronger bond.*

36 Others, however, indicated that improving technological capabilities allowed close relationships to
37 be maintained at a long distance including one (CDX13) whose mother was living abroad.

38 CDX13 [female, mother]: *...I think the beauty of our time is technology, so I can see her*
39 *through the PlayStation now, we've got a nice big screen in our living room and it's like we're*

1 *in the same room, she sits there and we show her the kids and we don't have to put the*
2 *Skype phone down or carry around a laptop so it's really easy and I think that distance, even*
3 *without her disease it's really helped us keep in touch all the time, I feel like whenever I want*
4 *to have a cup of tea with my mum I just turn on the TV and she's there, so I think in that*
5 *sense we've been really really lucky, I don't know if that would have happened about ten*
6 *years [ago].*

7 CDX16 [female, father]:...*you just pick up the phone...or Skype or everything else.*

8 CDX25 [female, father]:...*you know you can communicate in so many ways now, and just*
9 *talking on the phone even, you can have a heart to heart, you can still have that connection.*

10 4. Discussion

11 There has been debate within health economics regarding who should be included within economic
12 evaluation, with reference cases internationally suggesting those other than the patient should be
13 incorporated [5,6]. This paper takes a first step into exploring the close-person networks of those at
14 end of life. A novel hierarchical mapping approach was used to facilitate exploration. The median
15 number of close-persons was eight, with three in the inner (closest) ring. For the majority the
16 closest people were (unsurprisingly) family members. There was, however, significant heterogeneity
17 in network size.

18 Network sizes here were similar to those reported in other contexts [30] (8 vs 8.9), as was the size of
19 the inner ring in older populations elsewhere [29,30] (3 vs 3.5 confidantes/inner circle). This
20 research, supported by these other studies, suggests that it is the impacts upon these closest
21 (median) three individuals that economists may want to capture as a priority for economic
22 evaluation; this would certainly provide a starting point for capturing spill-over impacts. For a more
23 comprehensive societal perspective it may be desirable to capture wider impacts to the whole
24 network (median eight), although this may be of limited practicality in many research settings.

25 Non-family members featured in over 60% of the hierarchical maps, suggesting that it is important
26 to look beyond the family if all significant effects are not to be missed. Many participants felt that
27 geographical closeness was not necessary and some reported close relationships with overseas
28 decedents suggesting that those who do not reside in close proximity to the decedent may also need
29 to be included in economic evaluations. This may have practical implications for research conduct,
30 particularly regarding the jurisdiction of research ethics committees, rules around data protection
31 issues in different countries and whether impacts to those in other jurisdictions are relevant to local
32 decision makers.

33

34 The hierarchical mapping technique proved to be a strength within the interview process. It was a
35 simple and quick way to elicit data on potential networks for inclusion in economic evaluation.
36 Furthermore, it gave participants the time to reflect upon social networks at end of life, and the
37 relative strengths of their relationships as reported elsewhere [42]. This would have been difficult to
38 attain through solely verbal interaction. The hierarchical map also provided the interviewer with a
39 visual platform to probe the details of relationships, whilst also facilitating rapport, as in previous
40 research using pictorial tools [37,47].

41 The primary limitation of this research is the identity of the person completing the hierarchical
42 maps. 'Who' completed the task will inevitably shape perceptions of networks. Only one participant
43 was recruited via snowball sampling which meant that comparison across the same networks

1 derived from different informants, could not be achieved. Research within the wider social network
2 literature suggests that there are limits to the ability of individuals to identify the social networks of
3 others [48] although some work has found congruence between the principal and close-persons to
4 be high in terms of network membership, whilst varying by individual asked [49]. Spouses and family
5 members appear to be the most accurate respondents whilst friends were generally poor. Although
6 caution may be required in generalising the results found here [50], the logistical issues in capturing
7 data directly from decedents may preclude alternative approaches, particularly where decedents are
8 cognitively impaired before death. The sample in this study covered a wide variety of death
9 trajectories and health conditions. Specific nuances of the hierarchical map may also influence
10 network size, for example the size of circles or arrows may lead to participants setting out their
11 networks in a certain way. A further limitation of the hierarchal mapping process is that it will
12 inevitably exclude those who have no close-persons. Loneliness and social isolation is prevalent issue
13 among older people within the UK, with 10% of those aged over 65 years describing themselves as
14 often or always lonely [51] and around 11,000 funerals nationwide conducted without family or
15 friends being present [52].

16 There are many challenges in including wider impacts within economic evaluation. There are three
17 scenarios where equity concerns are particularly pertinent:

- 18 1. There is an implicit implication that cases with larger networks will accrue more benefits
19 than those with small networks, and thus will be prioritised over isolated individuals.
- 20 2. Interventions that target close persons as well as patients will be prioritised over
21 interventions with benefits solely to the patient.
- 22 3. Given death trajectory appears to impact network size, certain disease groups may be
23 disadvantaged by including close-person benefits within economic evaluation.

24 In practice, the first scenario is unlikely due to the analytic focus at group level. The second scenario
25 highlights the fundamental normative issue around who the health service is intended to benefit. If
26 we accept that we should be interested in capturing the wider benefits of treatments, then this is an
27 inevitable consequence. The third scenario highlights the issue of the point at which network size
28 should be considered. For example, for those with irreversible conditions affecting cognitive ability,
29 should the network size at the start of their disease or towards the end of life be the one that
30 'counts'? This scenario also highlights that some conditions may have disproportional impacts on
31 family members.

32 Whilst small steps towards including carer impacts within economic evaluation have been made
33 [53], including other 'close-persons' will create further practical challenges for researchers and
34 decision makers. These include: how to weight impacts for close persons relative to patients, how to
35 weight between close persons with different levels of closeness, and how to compare cost-
36 effectiveness between interventions where wider impacts are captured with those where they are
37 not. Future research is necessary to investigate the public's preferences for allocating resources
38 between the decedent and their close person.

39 Given the relatively early development of this area of research there are many avenues for further
40 study. First, adopting a hierarchical mapping procedure longitudinally through the death trajectory
41 would highlight how networks change as death approaches. This study has only sought to answer
42 the question of 'who is close' rather than that of 'who is impacted most'. There is an implicit
43 presumption that the greatest impacts fall on those closest to the dying person, but there may be
44 instances where this is not the case. By incorporating, in economic evaluations, a measure of end of
45 life and bereavement experience [9] for all those who are close it may be possible to further inform

1 this issue. Given the subjective nature of hierarchical mapping, future research should seek to
2 further examine the extent to which different network members see the network in similar ways
3 (akin to inter-rater reliability). This could be achieved through obtaining multiple hierarchical maps
4 from different close persons within the same network. There is scope to further improve consistency
5 of completion of the hierarchical maps. For example, the mapping process could be accompanied
6 with vignettes clarifying the degrees of closeness, whilst the impact of altering the design could be
7 explored.

8 Conclusion

9 Incorporating spill-over impacts within economic evaluation is challenging, with little guidance on
10 who should be included, and how we should identify those who are impacted. This paper is a first
11 step towards addressing these challenges within the end of life care context. These findings have
12 important implications for resource allocation.

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15 Conflicts of interest

16 Alastair Canaway has nothing to disclose. Hareth Al-Janabi has nothing to disclose. Philip Kinghorn
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18 Data availability

19 The individual level data generated during and/or analysed during this study are not publicly
20 available due to lack of consent for use in this way. Aggregated data are available from the
21 corresponding author on reasonable request

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23 AC was responsible for conducting the research and for the drafting of the manuscript. All authors
24 were involved in the development and design of the study. All authors contributed to the analysis of
25 the data. All authors reviewed, commented, and edited drafts of the manuscript.

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References

- [1] Drummond M, Sculpher M, Torrance G, O'Brien B, Stoddart G. *Methods for the Economic Evaluation of Health Care Programmes*. vol. Third Edit. Oxford: Oxford University Press; 2005.
- [2] NICE. *NICE Guide to the Methods of Technology Appraisal*. *Pharmacoeconomics* 2008;26:725–7. doi:10.2165/00019053-200826090-00002.
- [3] NICE. *Guide to the methods of technology appraisal 2013*. NICE; 2013.
- [4] NICE. *Guide to the Methods of Technology Appraisal*. NICE; 2004.
- [5] National Healthcare Institute. *Guideline for economic evaluations in healthcare*. 2016.
- [6] Sanders GD, Neumann PJ, Basu A, Brock DW, Feeny D, Krahn M, et al. Recommendations for Conduct, Methodological Practices, and Reporting of Cost-effectiveness Analyses. *JAMA* 2016;316:1093. doi:10.1001/jama.2016.12195.
- [7] Brouwer W, van Exel N, van Gorp B, Redekop W. The CarerQol instrument: a new instrument to measure care-related quality of life of informal caregivers for use in economic evaluations. *Qual Life Res Int J Qual Life Asp Treat Care Rehabil* 2006;15:1005–21. doi:10.1007/s11136-005-5994-6.
- [8] Brouwer WBF. Too Important to Ignore. *Pharmacoeconomics* 2006;24:39–41. doi:10.2165/00019053-200624010-00003.
- [9] Canaway A, Al-Janabi H, Kinghorn P, Bailey C, Coast J. Development of a measure (ICECAP-Close Person Measure) through qualitative methods to capture the benefits of end-of-life care to those close to the dying for use in economic evaluation. *Palliat Med* 2016;0269216316650616. doi:10.1177/0269216316650616.
- [10] Coast J. Strategies for the economic evaluation of end-of-life care: making a case for the capability approach. *Expert Rev Pharmacoecon Outcomes Res* 2014;14:473–82. doi:10.1586/14737167.2014.914436.
- [11] Coast J, Bailey C, Canaway A, Kinghorn P. Measuring and valuing outcomes for care at the end of life: The capability approach. 2016. doi:10.1007/978-3-319-28267-1_7.
- [12] Hoefman R, van Exel J, Brouwer W. Measuring the impact of caregiving on informal carers: a construct validation study of the CarerQol instrument. *Health Qual Life Outcomes* 2013;11:173. doi:10.1186/1477-7525-11-173.
- [13] Al-Janabi H, van Exel J, Brouwer W, Coast J. A Framework for Including Family Health Spillovers in Economic Evaluation. *Med Decis Mak Int J Soc Med Decis Mak* 2016;36:176–86. doi:10.1177/0272989X15605094.
- [14] Al-Janabi H, Flynn TN, Coast J. Estimation of a preference-based carer experience scale. *Med Decis Mak Int J Soc Med Decis Mak* 2011;31:458–68. doi:10.1177/0272989X10381280.
- [15] Coast J. Strategies for the economic evaluation of end-of-life care: making a case for the capability approach. *Expert Rev Pharmacoecon Outcomes Res* 2014;14:473–82. doi:10.1586/14737167.2014.914436.
- [16] Haycox A. Optimizing Decision Making and Resource Allocation in Palliative Care. *J Pain Symptom Manage* 2009;38:45–53.
- [17] Cameron J, Parkes CM. Terminal care: evaluation of effects on surviving family of care before and after bereavement. *Postgrad Med J* 1983;59:73–8.
- [18] Ransford HE, Smith ML. Grief resolution among the bereaved in hospice and hospital wards. *Soc Sci Med* 1991;32:295–304.
- [19] Schulz R, Mendelsohn AB, Haley WE, Mahoney D, Allen RS, Zhang S, et al. End-of-Life Care and the Effects of Bereavement on Family Caregivers of Persons with Dementia. *N Engl J Med* 2003;349:1936–42.
- [20] Christakis NA, Iwashyna TJ. The health impact of health care on families: a matched cohort study of hospice use by decedents and mortality outcomes in surviving, widowed spouses. *Soc Sci Med* 2003;57:465–75.
- [21] Stroebe M, Schut H, Stroebe W. Health outcomes of bereavement. *The Lancet* 2007;370:1960–73.

- 1 [22] Granovetter M. The Strength of Weak Ties. *Am J Sociol* 1973;78:1360 – 1380.
2 doi:10.2307/2776392.
- 3 [23] Marsden PV, Campbell KE. Reflections on Conceptualizing and Measuring Tie Strength. *Soc*
4 *Forces* 2012;91:17–23. doi:10.1093/sf/sos112.
- 5 [24] Petróczi A, Nepusz T, Bazsó F. Measuring tie-strength in virtual social networks. *Connections*
6 2007;27.2:39–52.
- 7 [25] Starzyk KB, Holden RR, Fabrigar LR, Macdonald TK. The Personal Acquaintance Measure: a tool
8 for appraising one’s acquaintance with any person. *J Pers Soc Psychol* 2006;90:833–47.
9 doi:10.1037/0022-3514.90.5.833.
- 10 [26] Aron A, Mashek D, Aron E. Handbook of Closeness and Intimacy. In: Mashek D, Aron A,
11 editors., New Jersey: Psychology Press; 2004, p. 27–30.
- 12 [27] Antonucci TC. Hierarchical mapping technique. *Gener J Am Soc ...* 1986;10:10–2.
- 13 [28] Moreno J. Who shall survive?: A new approach to the problem of human interrelations. 1934.
- 14 [29] Cornwell B, Schumm LP, Laumann EO, Graber J. Social Networks in the NSHAP Study: Rationale,
15 Measurement, and Preliminary Findings. *J Gerontol Ser B* 2009;64B:i47–55.
16 doi:10.1093/geronb/gbp042.
- 17 [30] Antonucci TC, Akiyama H. Social Networks in Adult Life and a Preliminary Examination of the
18 Convoy Model. *J Gerontol* 1987;42:519–27. doi:10.1093/geronj/42.5.519.
- 19 [31] Tomini F, Tomini SM, Groot W. Understanding the value of social networks in life satisfaction
20 of elderly people: a comparative study of 16 European countries using SHARE data. *BMC*
21 *Geriatr* 2016;16:203. doi:10.1186/s12877-016-0362-7.
- 22 [32] NHS Choices. Hospice care - End of life care guide - NHS Choices 2018.
23 <https://www.nhs.uk/Planners/end-of-life-care/Pages/hospice-care.aspx> (accessed January 16,
24 2018).
- 25 [33] Frank O, Snijders T. Estimating the Size of Hidden Populations Using Snowball Sampling. *J Off*
26 *Stat* 1994;10:53–67. doi:10.1214/aoms/1177705148.
- 27 [34] Ingleton C, Morgan J, Hughes P, Noble B, Evans A, Clark D. Carer satisfaction with end-of-life
28 care in Powys, Wales: a cross-sectional survey. *Health Soc Care Community* 2004;12:43–52.
29 doi:10.1111/j.1365-2524.2004.00467.x.
- 30 [35] Rempel GR, Neufeld A, Kushner KE. Interactive use of genograms and ecomaps in family
31 caregiving research. *J Fam Nurs* 2007;13:403–19. doi:10.1177/1074840707307917.
- 32 [36] McCormick KM, Stricklin S, Nowak TM, Rous B. Using Eco-Mapping to Understand Family
33 Strengths and Resources. *Young Except Child* 2008;11:17–28. doi:10.1177/1096250607311932.
- 34 [37] King N, Bravington A, Brooks J, Hardy B, Melvin J, Wilde D. The Pictor technique: a method for
35 exploring the experience of collaborative working. *Qual Health Res* 2013;23:1138–52.
36 doi:10.1177/1049732313495326.
- 37 [38] Antonucci TC, Akiyama H, Takahashi K. Attachment and close relationships across the life span.
38 *Attach Hum Dev* 2004;6:353–70. doi:10.1080/1461673042000303136.
- 39 [39] Antonucci TC, Akiyama H, Lansford J. Negative effects of close social relations. *Fam Relat*
40 1998;47:379–84.
- 41 [40] Hlebec V, Mrzel M, Kogovšek T. Social support network and received support at stressful
42 events. *Metodološki Zv* 2009;6:155–71.
- 43 [41] Santos JD, Levitt MJ. Intergenerational Relations with In-Laws in the Context of the Social
44 Convoy: Theoretical and Practical Implications. *J Soc Issues* 2007;63:827–43.
45 doi:10.1111/j.1540-4560.2007.00539.x.
- 46 [42] Canaway A, Al-Janabi H, Kinghorn P, Bailey C, Coast J. Incorporating novel qualitative methods
47 within health economics: the use of pictorial tools. *Qual. METHODS Health Econ., Rowman &*
48 *Littlefield*; 2017, p. 205–16.
- 49 [43] Glaser BG. The Constant Comparative Method of Qualitative Analysis. *Soc Probl* 1965;12:436–
50 445. doi:10.1525/sp.1965.12.4.03a00070.

- 1 [44] Ritchie J, Lewis J. Qualitative Research Practice: A Guide for Social Science Students and
2 Researchers. vol. 1. 2nd ed. London: SAGE Publications; 2013.
- 3 [45] Mays N, Pope C. Qualitative research in health care: Assessing quality in qualitative research.
4 BMJ 2000;320:50–2. doi:10.1136/bmj.320.7226.50.
- 5 [46] ONS. Deaths registered in England and Wales: 2017 2018.
6 [https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/deaths/
7 bulletins/deathsregistrationsummarytables/2017](https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/deaths/bulletins/deathsregistrationsummarytables/2017) (accessed July 30, 2018).
- 8 [47] Ray RA, Street AF. Ecomapping: an innovative research tool for nurses. J Adv Nurs
9 2005;50:545–52. doi:10.1111/j.1365-2648.2005.03434.x.
- 10 [48] Stansfeld S, Marmot M. Deriving a survey measure of social support: The reliability and validity
11 of the close persons questionnaire. Soc Sci Med 1992;35:1027–35. doi:10.1016/0277-
12 9536(92)90242-I.
- 13 [49] Antonucci TC, Israel B. Veridicality of social support: A comparison of principal and network
14 members' responses. J Consult Clin Psychol 1986;54:432–7. doi:10.1037/0022-006X.54.4.432.
- 15 [50] Marsden P V. Network Data and Measurement. Annu Rev 1990;16:435–63.
- 16 [51] Age UK. Loneliness - the state we're in 2012.
17 [http://www.ageuk.org.uk/brandpartnerglobal/oxfordshirevpp/documents/loneliness the state
18 we are in - report 2013.pdf](http://www.ageuk.org.uk/brandpartnerglobal/oxfordshirevpp/documents/loneliness%20the%20state%20we%20are%20in%20-%20report%202013.pdf).
- 19 [52] Burstow P. Dying Alone: Assessing isolation, loneliness and poverty 2005.
20 [http://image.guardian.co.uk/sys-
21 files/Society/documents/2006/04/07/Dying_Alone_Isolation_Report2final.doc](http://image.guardian.co.uk/sys-files/Society/documents/2006/04/07/Dying_Alone_Isolation_Report2final.doc).
- 22 [53] Hoefman RJ, Exel J van, Brouwer WBF. Measuring Care-Related Quality of Life of Caregivers for
23 Use in Economic Evaluations: CarerQol Tariffs for Australia, Germany, Sweden, UK, and US.
24 PharmacoEconomics 2017;35:469–78. doi:10.1007/s40273-016-0477-x.
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26 **Figures legend**

27 **Figure 1** Example of completed hierarchical map

28 **Figure 2** Hierarchical map for CDX8 – family are not always the closest
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3 **Volunteers required for study on** 4 **End-of-Life and Bereavement**

5
6 Have you been bereaved in the last 6-24 months and would feel comfortable discussing your
7 experience with a researcher? Alternatively, is somebody close to you currently receiving end of life
8 care? If so, then we would like to invite you to participate in our study investigating how end-of-life
9 impacts family and friends.

10 The study aims to improve the evaluation of end of life care in the UK. Confidentiality and sensitivity
11 will be guaranteed. You will be sent additional information and be able to discuss the study with the
12 researcher before being asked to decide whether or not to participate. For more information email
13 Alastair Canaway at axc105@bham.ac.uk

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1 Appendix 2: Hierarchical Mapping Template

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