

Family caregiver participation in palliative care research: Challenging the myth

Authors:

Professor Samar Aoun¹, Professor of Palliative Care

Dr Susan Slatyer^{1,2}, Research Fellow

Kathleen Deas¹, Research Officer

Dr. Cheryl Nekolaichuk³, Professor, Palliative Care Medicine

Email: s.aoun@curtin.edu.au

Email: s.slatyer@curtin.edu.au

Email: kathleendeas@optusnet.com.au

Email: Cheryl.Nekolaichuk@covenanthealth.ca

¹School of Nursing, Midwifery and Paramedicine, Curtin University, GPO Box U1987, Perth WA 6845, Australia

²Centre for Nursing Research, Sir Charles Gairdner Hospital, Harry Perkins Institute for Medical Research, Hospital Ave, NEDLANDS WA 6009

³Division of Palliative Care Medicine, Department of Oncology, University of Alberta, 1090 Youville Drive West Edmonton, AB T6L 5X8

Corresponding author:

Samar Aoun

Curtin University, GPO Box U1987, Perth, WA 6845, Australia

Telephone: +61 419 911 940

Email: s.aoun@curtin.edu.au

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Abstract

Background: Despite international guidelines emphasizing consumer directed care and autonomous decisions in research participation, there is a common myth that research can be an additional and unwanted burden on patients and their family members.

Aim: to examine the experiences and impact of research involvement on family caregivers (FC) of terminally ill people, focusing within home-based palliative care.

Design and participants: 316 of 322 participants (98.1%), who completed a FC support intervention through a stepped wedge cluster trial (Australia, 2012-15), participated in a post-intervention telephone interview on their study experiences, which included quantitative and qualitative questions.

Results: 97% of both the control (n=89) and intervention (n=227) groups perceived positive aspects, while almost all did not report any negative aspects of being involved in this research; the majority rated their involvement as very/extremely beneficial (control 77%; intervention 83%). The qualitative analysis generated three major themes: "Intrapersonal – Inward directed"; "Connection with others – Outward directed"; and "Interpersonal – Participant researcher relationship".

Conclusions: This study provided quantitative and qualitative evidence challenging the myth. In contrast to health professional concerns, FCs appreciated the opportunity to participate and benefited from their involvement in research. Research protocols need to be specifically tailored to the needs of family caregivers and include debriefing opportunities for all participants at the end of interventions studies, regardless of which group they have been assigned. Strategies that facilitate health professionals' understanding of the research and risk-benefits may help reduce gatekeeping and improve the validity of research findings.

Keywords: Palliative care research, gatekeeping, research participation, family caregivers.

Introduction

There is a need to improve the evidence base in palliative care, using patient and family participants, to inform clinical decision-making and service provision for palliative care patients and their family caregivers (1-4). Without the perspectives of patients and their family caregivers, it is difficult to understand their needs and develop interventions to meet these needs. Indeed, patients see research as the key to improving care (5). Moreover, when asked, patients at end-of-life both envisage and report benefit from participating in research (6-8).

A number of national and international guidelines have emphasized the importance of acknowledging the patients' and family caregivers' needs and wishes to guide decision-making

and care planning (9-12). The American Society of Clinical Oncology has further advocated for patients to be given the opportunity to participate in clinical trials or other forms of research (13). More recently, the MORECare recommendations for ethical issues in palliative and end-of-life (EOL) research have included the need for “autonomous decisions of patients and caregivers regarding their participation in research to avoid limiting their participation through inappropriate gatekeeping and paternalistic attitudes” (14).

The benefits of patients’ participation in research studies are well documented in the literature (5-7, 15, 16). Alexander (15) found that patients in the last months of life were overwhelmingly “eager to participate” (15p.174). Striking evidence that patients even very close to death can draw positives from research involvement emerged from a study conducted in an Australian hospice (5). For some patients, the research offered a way to do something valuable with the little time that remained, while for others participation provided validation of their own worth and life “outside of being the dying person” (5, p.408). In valuing knowledge that had informed their own care and treatment, many patients sought to contribute to benefit others. Lung cancer patients who were participating in a clinical trial identified a number of perceived benefits such as treatment benefit, altruism, personal fulfilment and positive attention (16). In a study by Pessin et al (17), the majority of patients reported no burden associated with participation (75%) and found the experience as moderately to highly beneficial (68%). Factors most frequently identified as beneficial were the social interaction (75%), sense of contributing to society (57%), and the opportunity to discuss their illness (47%).

However less is known about the family caregivers’ experiences of research participation. The stress of caring for a dying relative is well recognized and includes fatigue, emotional distress, diminished quality of life, and social restriction and isolation (18-20). Despite such concerns, there is some evidence that family caregivers can experience similar benefits from research participation as do patients (21, 22).

Aoun and Nekolaichuk (1) recently challenged reported concerns that research participation is neither well-tolerated nor wanted by palliative care patients and their families. They drew on evidence that patients and families wished to decide for themselves whether or not to contribute to studies, and that they experienced benefits, such as personal gain and a sense of altruism, when they did so (17, 23). Accordingly, these authors considered participant burden to be one of five myths about palliative care research that could limit the quantity and quality of investigations. As has been recognized elsewhere, when patients and family caregivers are deemed suitable for participation on the basis of staff members’ perceptions, rather than ethically approved inclusion and exclusion criteria, the representativeness of study samples and, ultimately, the research merit can be limited (1, 24, 25).

Objective

The purpose of this study was to examine the experiences and impact of research involvement on family caregivers of terminally ill people in home-based palliative care, by focusing on both the positive and negative aspects of study participation. Unlike previous studies on this topic,

this study included the experiences of participants assigned to either an intervention or control group.

Methods

The study was approved by Curtin University Human Research Ethics Committee (HR 24/2011) and Silver Chain Human Research Ethics Committee (EC App 068).

The sample consisted of 322 primary family caregivers of palliative care patients, who were receiving home-based palliative care services through Silver Chain Hospice Care Service, in Perth, Western Australia. Participants had been involved in one of two arms of a stepped wedge cluster trial to implement the Carer Support Needs Assessment Tool (CSNAT), with an intervention group using the CSNAT for identification of support needs and a control group using the service standard identification of support needs (26, 27). Recruitment at baseline was 620. However, there was 45% attrition across both groups between baseline and follow-up, mainly due to patient deaths, resulting in 322 caregivers completing the study (233 in the intervention group and 89 in the control group). The intervention period spanned a period of 2-3 weeks (2012-15). The study design, description of the intervention, outcome measures collected at pre and post intervention and results of the trial are described in detail in (26).

This article reports on one component of the post intervention data collection, which is the feedback about participation in research, undertaken a week after the intervention was completed. This consisted of a brief three-item questionnaire, adapted from Hudson et al (21), focusing on the experience of family caregivers participating in this research. The first two questions related to whether participants perceived any positive or negative aspects undertaking this research (response yes or no). The third question consisted of a five-point Likert scale rating the extent of the benefit of being involved in the research (0 = not at all beneficial, 1 = somewhat beneficial, 2 = not sure, 3 = very beneficial, 4 = extremely beneficial). Each of the three questions provided participants with opportunities for comments to elaborate on their responses.

This study adopted a mixed methods design, incorporating quantitative and qualitative methods. Brief telephone interviews took place with all 322 participants who completed the study, at a pre-arranged time convenient to participants. Interviews from the first 80 control and intervention participants were audio-recorded, transcribed verbatim and then analyzed, until no new themes arose from the conversations i.e. data saturation was reached. For the remaining interviews, the interviewer took detailed notes to validate emerging findings from the first 80 interviews. Some participants spoke in detail about their experiences when probed. For others, responses were limited to yes/no with a few additional comments.

Analysis

Data from the interviews were subjected to a thematic analysis (28, 29). Initial coding was carried out independently by two co-authors, one of them being the interviewer so that consideration of the nonverbal context was assured (30). Coding was supported by the NVivo 10 software (31). Transcriptions were read and re-read to identify key words and phrases that

were then grouped into categories labeled with codes. Themes emerging after comparisons within and among individual interviews identified key messages, representing caregivers' verbal descriptions. Two co-authors independently identified these themes initially, with differences resolved by discussion and returning to the data. The explanation of themes and how interpretations had been reached were supported by exemplars, consisting of family caregiver quotes (29). To further ensure the trustworthiness of our findings (30), transferability was established by our comprehensive description of the study's setting and participants. Descriptive statistics, using SPSS v.22, were used for analyzing quantitative data.

Quantitative Results

316 out of the 322 participants completed the post-intervention feedback interview, focusing on their experiences with research participation (response rate 98.1%). As the following findings pertain to just one component of the post-intervention data collection, it was not possible to separate the exact time spent on just this component. On the whole, the median duration of telephone interviews was 19 minutes ranging from 9 to 63 minutes.

Profile of participants

Table 1 displays a comparison of baseline demographic data between the intervention (n=227) and control (n=89) groups, for those participants who completed the study. Caregivers did not differ significantly in most characteristics, except in the following: intervention participants were significantly younger ($p = 0.037$) and their caring role affected their work ($p=0.008$); they also had more care recipients with cancer diagnoses ($p = 0.039$), shorter median lengths of stay (LOS: length of time patients were registered with the service as clients) with Silver Chain ($p=0.001$) and shorter median LOS in palliative care ($p < 0.001$) than control participants.

[insert Table 1 about here]

Ratings of experience

Ninety-seven percent of both the control and intervention groups perceived positive aspects to being involved in this research. 100% of the control group and 98% of the intervention group did not report any negative aspects to their participation in research. Figure 1 shows the extent to which the participants perceived the research was beneficial. The overwhelming majority in both groups rated their involvement in this research as very or extremely beneficial (control 77% and intervention 83%).

[Insert Figure 1 about here]

Fig 1: Participants' ratings of benefit in research participation, for both control and intervention groups (n=316).

Qualitative Results

The qualitative analysis generated three major themes describing the benefits that participants reported in response to participating in the research: (a) “Intrapersonal – inward directed”; (b) “Interpersonal – Connection with others”; and (c) “Interpersonal – Participant researcher relationship”. Table 2 displays these themes and the corresponding sub-themes.

[Insert Table 2 about here]

Theme 1: “Intrapersonal – Inward directed”

This theme related to the feelings and thoughts that caregivers experienced as a result of their participation in this research. The majority of participants in both groups reported receiving benefit, with many deriving comfort from feeling valued and cared for. A typical comment was, *“Being able to take part has been a privilege to be involved in something like this [research]. Thank you for being so compassionate”* (Intervention 136). Three sub-themes were generated that indicated how participants’ involvement in the study offered them time for reflection; increased their insight into their own emotional concerns and provided a sense of validation that reassured them in their caregiving role. These sub-themes are briefly described.

Prompting contemplation and reflection: “Making you think”

There was a sense from both groups that the process of data collection gave participants an opportunity to ‘take stock’, and reflect. Interaction with the research nurse could prompt caregivers to consider the impact of the caregiver role, as one participant said: *“After you read through the list of questions it makes you think about other things. You often do things without thinking, and doing this [research] makes you realise what you actually do [as a carer]”* (Control 80). In particular, completing the questionnaires tended to crystallize the emotional impact of caring for a loved one who was seriously ill, *“It makes you think a little – made me realise the emotional side of caring and how [with] a lot of those issues you tend to ‘soldier on’”* (Intervention 115). Moreover, as caregivers were prompted to reflect about the issues at hand, they developed greater insight into both their own need for support and the supports available to them.

Gaining insight and awareness of support needs: Recognizing “the emotional and physical side of caring”

Caregivers in both the intervention and control groups described how working through the various questionnaires helped to illuminate the challenges they faced. For example, one participant remarked, *“It gives more awareness for caregivers – the emotional and physical side of caring”* (Control 51). Another stated, *“It’s making me think – made me bring things out I wasn’t aware that . . . [I was] feeling. I may have been denying, but it has helped me doing this. It definitely helped me”* (Control 57).

Feeling acknowledged and valued: “Feeling as if you matter”

There was a general appreciation of this research initiative that acknowledged the caring role and valued the caregivers’ perspectives. For example, one caregiver remarked, *“It recognises the carer and what they are doing. Fantastic for someone to be doing this research”* (Control

48), while another appreciated that the research *“acknowledges all the things that are going on with your life when you become a carer. So it reaffirms what you are doing and does help”* (Intervention 332).

Participation in the research seemed to validate the caregivers’ perspectives, which so often seemed to go unrecognised. For example, two caregivers declared, *“Any research in this subject is very good. Friends don’t realise all the tasks required when you become a carer”* (Control 47); and, *“Reading the pamphlet helped – makes you aware of what’s involved in being a carer. A positive result, my sister now realises how much support my wife needs”* (Control 89). As a consequence, one caregiver reported: *“A psychological benefit in knowing that someone is thinking about these issues actually helped me. At least someone is doing research in this area and may make some changes to help others in our situation”* (Control 128). In turn, the insight gained through the research interactions was reassuring, as one participant reported: *“Taking time to think about things – about you as a caregiver, makes you feel as if you matter as well. Self-awareness of what you are actually doing”* (Intervention 107).

Theme 2: “Connection with others – Outward directed”

Knowing that someone cared about caregivers was helpful to these participants. Additionally, it was evident that participants were helped by perceptions that they could help others who potentially faced the burden and isolation of caregiving. A sense that ‘helping others helps me’ emerged as evidenced by the following participant’s comment: *It’s important these things are done. You have to get information to make any improvements. It’s beneficial to me to be part of this. It gladdens me – it’s good to be able to help others in the future.* (Control 219). Three sub-themes were generated:

Feeling less isolated: “Letting people in”

Participants in this study indicated that the additional burden they shouldered as caregivers was not always visible to others. This stemmed, in part, from a reluctance to share problems as evidenced by one caregiver’s recollection who said, *“At first I tried to do everything alone and it felt at first that I’d failed if I couldn’t do everything”* (Control 32). For some participants, the outcome was a sense of isolation, as this caregiver explained, *“People can feel very isolated as a carer – you’re separated from society and only in your caring role”* (Intervention 474). For these caregivers, involvement in the research process offered the opportunity to share their experiences and feelings. Caregivers reported that this tended to help them feel more connected to others.

Regardless of the group allocation, interaction with the research nurse tended to reduce participants’ feeling of isolation, as described by the following participants’ quotes:

“Someone’s there to listen – that’s very important because it can get very lonely as a carer” (Intervention 536)

“Thinking about the questions asked, making me more aware (as a first time carer). . . It has been a benefit to me. I know that I am not totally alone in this as the cancer progresses” (Control 32).

“It was nice to talk to someone. It makes you feel part of everything instead of just being a carer and looking after someone. It involves you completely” (Intervention 339).

When caregivers were supported to reflect on their needs, some were prompted to seek resources to cope, as illustrated by the following quotes:

“It made me realise that I can’t do this on my own and rang up to get help” (Intervention 196).

“Now I’ve learnt from this [involvement in the research] and I’ve now ‘let people in’ to help” (Control 32).

Improving support for future caregivers: “Making a difference for others”

It was evident that participants believed their involvement contributed to improving support for future caregivers and influencing service improvement. Motivation by a sense of altruism to participate in the research was demonstrated by participants from both groups; *“The whole reason for me doing it was to help other people. There’s always room for improvement – its why these things [research] are done”* (Intervention 206).

Both control and intervention group participants indicated the research findings provided an opportunity to articulate their experiences and challenges, giving caregivers a voice. One participant expressed, *“If people are made aware of the feelings caregivers are going through, it may be beneficial in providing more support for others”* (Control 17). Resonating with the altruistic motives of others, one caregiver stated, *“I am happy to do whatever I can to help if it makes a difference for others. Unless the government knows the details of what we need they can’t make a difference”* (Control 77).

Caregivers allocated to the control group did not receive the intervention but valued their participation nonetheless. As one caregiver from the control group explained, *“I’m willing to help anybody. It hasn’t upset me. I’m going through every stage anyway and talking about it hasn’t upset me. If it helps someone else it will be good”* (Control 86). Participants expected that much of this perceived future benefit would be achieved through service improvement.

Enhancing service improvement: “Needing documented evidence”

The chance to advocate for the organisation was seen as important, as illustrated by one of the participants: *“Without research you can’t have change. In research, documented evidence is needed for funding. If [they, the organisation] want changes and funding, they need documented evidence”* (Intervention 515).

Some participants, however, were more circumspect about whether or not the research would bring expected improvements. On a personal level, one participant realised that she was early in her journey as a caregiver, declaring, *“I’m only halfway caring for my husband so I can’t make a judgement yet”* (Control 177).

Other participants tended to want more evidence regarding service improvement before committing to a view. Accordingly, when asked about benefits from their involvement, several participants provided tentative or conditional responses: *I don’t know. [I’ll] have to wait and see*

what the outcome is (Intervention 529); I wouldn't know until anything is put in place (Intervention 329); It might help others as long as it goes through and something gets done (Control 232); "I think all research is good provided it has an outcome (Intervention 428).

Despite these reservations, participants embraced the opportunity to participate in this research. It seemed that any perceived risks of participation were outweighed by potential benefits, as one participant said, *"Yet to be determined of the overall impact but I agreed to take part in the hope this research might help someone in the future"* (Control 83).

Theme 3: "Interpersonal – Participant-researcher relationship"

The third theme described the benefits that participants reported from interacting with the researcher. Three sub-themes that described participants' experiences of their relationship with the researcher emerged: (a) "expressing a caring tone", (b) "listening to my heart" and (c) "respecting my free will."

Expressing a caring tone

The "multiple encounter telephone interview" research design enhanced rapport building, thus allowing the caregivers to feel safe in expressing their feelings to the interviewer. A compassionate and sincere interview style was important to maintain this relationship, as described by one participant: *"When you are talking to the carer, your disposition and tone of voice is important - the caring tone shows genuine interest and is a good thing for the caregiver - they pick up on this"* (Intervention 605).

Listening to my heart

The research experience, and specifically, the relationship with the researcher, provided an opportunity for caregivers to safely express their thoughts and feelings. In some cases, a new found confidence allowed the caregivers to confide their experiences with the researcher when previously unable to, *"It's good to be able to speak to someone about how you feel. I guess carers don't want to admit how they feel"* (Control 6). These interviews could enable caregivers to air their concerns and give voice to deeply held thoughts as evidenced by one participant who poignantly described interactions with the research assistant as: *"...an opportunity to have someone listen to my heart"* (Control 76).

Respecting my free will

Caregivers experienced their relationship with the researcher as a safe and impartial place to express themselves, as one participant declared, *"I can tell you and you are impartial with my answers. I think it was very beneficial"* (Control 8). It was evident that in this "safe place" caregivers felt comfortable to articulate their need for support, *"Just saying it out loud and having yourself saying it, helps in knowing where you are at"* (Control 25). The study used a flexible approach to ensure participants' priorities were respected, ensuring data collection did not increase caregiver's already considerable burden and that their participation was entirely voluntary., as shown in these two quotes, *"I didn't feel compelled to participate, no pressures to answer in a particular way"* (Intervention 146); *"It was totally my free will to take part"* (Intervention 193).

Discussion and Conclusion

To our knowledge, this study is a first in involving a cluster randomised large sample of family caregivers (n=316) in telephone interviews regarding their experiences in research participation (both quantitative and qualitative), as well as ascertaining the experiences of both the intervention and control groups rather than just an intervention group. Most caregiver intervention investigations have developed from descriptive studies using convenience samples (32). Both family caregiver groups appreciated the opportunity to be involved in this research project. Despite not receiving the intervention (just completing the outcome measures), the experiences of the control group participants were as positive as the intervention group. The therapeutic benefits of the intervention, which were specific just to the intervention group, were reported in a previous article (27).

From an inward directed (intrapersonal) perspective, participants' involvement in this study provided them with an opportunity to share their experiences, prompting reflection, as well as having increased insight into emotional concerns and a sense of validation in their role as a caregiver. Recognition of the importance of their self-worth can facilitate family caregivers recruitment and retention in research as reported by Murphy et al (32). While health professionals might seek to protect those they see as vulnerable, the denial of patients and their family members the opportunity to decide for themselves whether or not to participate in research violates the ethical principle of respect for autonomy (25, 33). It is apparent in this study that the ability of family caregivers to participate in research had been empowering and respectful of their autonomy.

Within the inward directed theme, the subthemes of "prompting contemplation and reflection" and "gaining insight and awareness of support needs" may provide family caregivers with considerable benefits. The lack of role identification amongst family caregivers and their reluctance to consider their own needs and seek help has been well-described in the literature (34, 35). The opportunity for participants to reflect and identify their support needs, within the research context, suggest that research participation, in itself, may help address these important issues, regardless of whether or not people receive the intervention. Therefore, deriving these benefits from the research experience may have introduced a confounding effect on the trial results, in so far as the reflection and identification of support needs prompted by the research activities were similar to the intended outcomes of the implementation of the CSNAT in the intervention arm. However, despite both groups being exposed equally to the same research activities, apart from the intervention, the intervention group was associated with a significant reduction in caregiver strain (26).

From an outward directed (connection with others) perspective, the findings indicated that the participants' opportunity to articulate their needs for support reduced the sense of isolation many caregivers were experiencing. In addition to helping themselves, participants also identified the ability to help others as a benefit of research participation. This sense of altruism has emerged in a number of palliative care studies (7, 15, 32, 36, 37). Caregivers may be more

willing to participate in intervention studies when they realize that the benefits may have multiple purposes that go beyond their personal needs (32), if their altruistic motivations are reinforced, and they understand the importance of the study. It is apparent that caregivers in this study perceived such altruistic benefits in terms of improving resourcing for other caregivers and influencing service improvement.

The findings from this study further emphasize the therapeutic benefit of the participant-researcher relationship. The qualitative approach facilitated the creation of relationships between participants and the research nurse that fostered respect, trust, and concern (38). The validation of the caregivers' emotions started at the initial contact and having the opportunity to discuss their concerns with the research nurse aided the study participant retention process (38). Caregivers may offer resistance to standardised tools, due to time constraints and disruptions in routines, desiring rather to tell more of their story (32). In this study, obtaining feedback by telephone and, as such, having a voice at the end of the line, was comforting and encouraged study participation and completion, in comparison to using self-administered questionnaires. Further, participants in our study described the researcher's approach for eliciting information from them as sensitive, being respectful of their time restrictions and overwhelming daily tasks, which is supported in the literature (38). The development of this relationship over the course of the study further facilitated participant retention.

Despite the lack of studies focusing on family caregivers' experiences of research participation, a number of parallels can be drawn between our study findings and studies focusing on patients' experiences of research participation. Similar to our findings, beneficial patient themes of validating self-worth (5), altruistic benefits of contributing to others or society (16, 17), and social interaction (17) have been identified in the literature. There are also some differences, however, between the family caregiver experiences in our study and patient research experiences in other studies, which may be due, in part to the type of study design. Drawing on the patient experiences that differed between the intervention and control groups in a randomised trial, Harrop et al (16) reported how control group participants reacted to (individual level) randomisation and came to terms with their allocation to the control arm, and identified alternative reasons and benefits for participation to those of the intervention group participants. These differences between the two groups did not feature in our study possibly due to the different study design. The stepped wedge cluster design used in our study is recommended where there is a firm belief in the benefits of the intervention, where costs are low and side effects are improbable, and where all participating sites were able eventually to undertake the intervention thus averting the disengagement within control sites (39). An implication for further research in this area would be to explore potential differences in caregiver experiences between types of study design.

Limitations

One of the limitations of this study was the unbalanced groups, with a higher proportion of intervention than control participants participating in the trial and therefore in the post-study interview regarding their research experiences. The issue of unbalanced groups is an inherent limitation of the stepped-wedge design. For the comparison of the two trial arms, however, the

generalized linear mixed modelling (GLMM) is generally robust to unbalanced groups, as explained in the trial article (26).

A second limitation is related to the attrition rate. Considering the 45% attrition rate in the trial, participants' views on research participation may have differed from those who could not complete the research. However, this attrition was predominantly due to patient deaths in both groups (Intervention 79.4%, Control 91.9%) and not because family carers did not have time or did not want to do the intervention (26). Compared to drop-outs, participants who completed the trial had a longer period of caring, were less likely to be in paid employment, and more likely to be from a non-English background. All other carer demographic variables were equivalent and patient profiles did not differ (26).

The research interviewer's existing relationship with the participants may be considered a third limitation. The researcher who conducted these interviews was part of the original research and thus already knew the people she was interviewing. This may have potentially caused some bias though equally for the two groups. There was no in-depth exploration of the lived experiences of participants in this study as this was not practical nor economical when the sample size was large (n=316).

Another limitation is that not all caregivers would have been approached by the nurses to participate, which may have given rise to a systematic selection bias. While gatekeeping cannot be ruled out, it may be that the decision to include a caregiver in the study would have been most likely influenced by the workload of the nurse on the day and how time-pressured she/he felt fitting in a number of patients who needed to be visited (26).

Finally, the implementation of the intervention happened in a single organization (although with three different geographical sites of the metropolitan area), which may affect generalizability.

Conclusions

In conclusion, as recommended by several international studies and literature reviews, (7, 14, 40), participants' perspectives concerning research participation is vital to acquire a thorough understanding of their experiences. This study provides further quantitative and qualitative evidence challenging the myth that research can be an additional and unwanted burden on patients and their family members (1). However, family caregivers may hold positive or negative attitudes towards participation in research and, therefore, interventions need to be uniquely designed so that caregivers can see that their investment will be worthwhile. Researchers need to balance maintaining a low burden for caregivers against administering a battery of tools to collect caregivers' experiences and needs. In addition, research protocols need to include debriefing opportunities for all participants, regardless of which group they have been assigned. Ongoing engagement and provision of information to facilitate health professionals' understanding of the research and risk-benefits may help reduce participant gatekeeping and thus improve the validity of research findings.

Competing interests

The authors declare that no competing interests exist.

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References

1. Aoun SM, Nekolaichuk C. Improving the evidence base in palliative care to inform practice and policy: thinking outside the box. *J Pain Symptom Manage.* 2014;48(6):1222-35.
2. Bennett MI, Davies EA, Higginson IJ. Delivering research in end-of-life care: problems, pitfalls and future priorities. *Palliat Med.* 2010;24(5):456-61.
3. Hui D, Arthur J, Dalal S, Bruera E. Quality of the supportive and palliative oncology literature: a focused analysis on randomized controlled trials. *Support Care Cancer.* 2012;20(8):1779-85.
4. Nambisan V. Research in palliative care -- a four-principle justification. *European Journal of Palliative Care.* 2010;17(2):76-9.
5. Terry W, Olson LG, Ravenscroft P, Wilss L, Boulton-Lewis G. Hospice patients' views on research in palliative care. *Intern Med J.* 2006;36.
6. Phipps E, Harris D, Braitman LE, Tester W, Thompson N, True G. Who enrolls in observational End of Life research? Report from the cultural variations in approaches to End of Life study. *J Palliat Med.* 2005;8.
7. Ross C, Cornbleet M. Attitudes of patients and staff to research in a specialist palliative care unit. *Palliat Med.* 2003;17(6):491-7.
8. Wilkinson A, Slatyer S, McCullough K, Williams A. Exploring the quality of life at the end of life (QUAL-E) instrument with Australian palliative care hospital patients: hurdles and directions. *J Palliat Care.* 2014;30(1):16-23.
9. Australian Commission on Safety and Quality in Health Care. Patient centred care: improving quality and safety through partnerships with patients and consumers. Sydney.: ACSQHC, 2011.
10. Australian Commission on Safety and Quality in Health Care (ACSQHC). National safety and quality health service standards. Sydney: ACSQHC, 2011.
11. Institute of Medicine. Crossing the quality chasm: a new health system for the twenty-first century. Washington National Academies Press; 2001.
12. National Institute of Clinical Excellence. Guidance on cancer services improving supportive and palliative care for adults with cancer. Oxford: National Institute of Clinical Excellence: National Institute of Clinical Excellence, 2004.
13. Peppercorn JM, Smith TJ, Helft PR, DeBono DJ, Berry SR, Wollins DS, et al. American Society of Clinical Oncology statement: toward individualized care for patients with advanced cancer. *J Clin Oncol.* 2011;29(6):755-60.

14. Gysels M, Evans CJ, Lewis P, Speck P, Benalia H, Preston NJ, et al. MORECare research methods guidance development: recommendations for ethical issues in palliative and end-of-life care research. *Palliat Med*. 2013;27(10):908-17.
15. Alexander SJ. 'As long as it helps somebody': why vulnerable people participate in research. *Int J Palliat Nurs*. 2010;16(4):174-9.
16. Harrop E, Noble S, Edwards M, Sivell S, Moore B, Nelson A. "I didn't really understand it, I just thought it'd help": exploring the motivations, understandings and experiences of patients with advanced lung cancer participating in a non-placebo clinical IMP trial. *Trials*. 2016;17(1):329.
17. Pessin H, Galietta M, Nelson CJ, Brescia R, Rosenfeld B, Breitbart W. Burden and benefit of psychosocial research at the end of life. *J Palliat Med*. 2008;11.
18. Aoun SM, Kristjanson LJ, Currow DC, Hudson PL. Caregiving for the terminally ill: at what cost? *Palliat Med*. 2005;19(7):551-5.
19. Lee S, Kristjanson L. Human research ethics committees: issues in palliative care research. *Int J Palliat Nurs*. 2003;9(1):13-8.
20. McMillan SC. Interventions to facilitate family caregiving at the end of life. *J Palliat Med*. 2005;8 Suppl 1:S132-9.
21. Hudson P. The experience of research participation for family caregivers of palliative care cancer patients. *Int J Palliat Nurs*. 2003;9(3):120-3.
22. Koffman J, Higginson IJ, Hall S, Riley J, McCrone P, Gomes B. Bereaved relatives' views about participating in cancer research. *Palliat Med*. 2012;26(4):379-83.
23. White C, Hardy J. What do palliative care patients and their relatives think about research in palliative care? - A systematic review. *Support Care Cancer*. 2010;18(8):905-11.
24. Tan H, Wilson A, Olver I, Barton C. Recruiting palliative patients for a large qualitative study: some ethical considerations and staff dilemmas. *Explore (NY)*. 2010;6(3):159-65.
25. Hudson P, Aranda S, Kristjanson LJ, Quinn K. Minimising gate-keeping in palliative care research. *European Journal of Palliative Care*. 2005;12(4):165-9.
26. Aoun S, Grande G, Howting D, Deas K, Toye C, Troeung L, et al. The impact of the Carer Support Needs Assessment Tool (CSNAT) in community palliative care using a stepped wedge cluster trial. *PLoS One*. 2015;10(4).
27. Aoun S, Deas K, Toye C, Ewing G, Grande G, Stajduhar K. Supporting family caregivers to identify their own needs in end-of-life care: qualitative findings from a stepped wedge cluster trial. *Palliat Med*. 2015:1-10.
28. Guest G, MacQueen KM, & Namey EE. *Applied thematic analysis*. London: Sage; 2012.
29. Hsieh H-F, Shannon SE. Three approaches to qualitative content analysis. *Qual Health Res*. 2005;15(9):1277-88.
30. Creswell JW. *Qualitative Inquiry and research design: choosing among five perspectives*. Thousand Oaks, CA: London: Sage publications; 2007.
31. QSR International Pty Ltd. NVivo qualitative data analysis software. 10 ed: QSR International Pty Ltd; 2012.
32. Murphy MR, Escamilla MI, Blackwell PH, Lucke KT, Miner-Williams D, Shaw V, et al. Assessment of caregivers' willingness to participate in an intervention research study. *Res Nurs Health*. 2007;30(3):347-55.
33. Sharkey K, Savulescu J, Aranda S, Schofield P. Clinician gate-keeping in clinical research is not ethically defensible: An analysis. *J Med Ethics*. 2010;36(6):363-6.

34. Harding R, Higginson I. Working with ambivalence: informal caregivers of patients at the end of life. *Support Care Cancer*. 2001;9(8):642-5.
35. Harrop E, Byrne A, Nelson A. "It's alright to ask for help": findings from a qualitative study exploring the information and support needs of family carers at the end of life. *BMC Palliat Care*. 2014;13:22.
36. Gysels M, Shipman C, Higginson I. Is the qualitative research interview an acceptable medium for research with palliative care patients and carers? *BMC Med Ethics*. 2008;9(1):7.
37. Whiting LS, Vickers PS. Conducting qualitative research with palliative care patients: applying Hammick's research ethics wheel. *Int J Palliat Nurs*. 2010;16(2):58, 60-2, 4-6, 8.
38. Steinhauser KE, Clipp EC, Hays JC, Olsen M, Arnold R, Christakis NA, et al. Identifying, recruiting, and retaining seriously-ill patients and their caregivers in longitudinal research. *Palliat Med*. 2006;20(8):745-54.
39. Brown CA, Lilford RJ. The stepped wedge trial design: a systematic review. *BMC Med Res Methodol*. 2006;6:54.
40. Kars MC, van Thiel GJ, van der Graaf R, Moors M, de Graeff A, van Delden JJ. A systematic review of reasons for gatekeeping in palliative care research. *Palliat Med*. 2016;30(6):533-48.

Family caregiver participation in palliative care research: challenging the myth

Tables

Table 1: Characteristics of groups who completed the CSNAT study and the research participation questionnaire (n=316)

		Control		Intervention		p-value
		N=89		N=227		
FAMILY CAREGIVER		n	%	n	%	
Gender						0.120
	Male	18	20.2	67	29.5	
	Female	71	79.8	160	70.5	
Age (yrs.)						0.037
	Mean ± SD	65.5 ±13.2		62.2 ±12.4		
	Median (Range min., max.)	67.0(33, 92)		62.0(20, 88)		
Marital status						0.173
	Never married	2	2.2	13	5.7	
	Widowed	2	2.2	6	2.6	
	Divorced/separated	9	10.1	10	4.4	
	Married/de facto	76	85.4	198	87.2	
Cultural background						0.121
	Australian	60	67.4	126	55.5	
	Other English speaking	21	23.6	65	28.6	
	Non-English speaking	8	9.0	36	15.9	
Usual employment						0.171
	Paid employment	17	19.1	71	31.3	
	Pensioner	44	49.4	93	41.0	
	Self-funded retiree	15	16.9	35	15.4	
	Other	13	14.6	28	12.3	
Education						0.615
	Primary	1	1.1	5	2.2	
	Secondary	57	64.0	132	58.1	
	Tertiary	31	34.8	90	39.6	
Living arrangements						0.164
	Private residence	81	91.0	217	95.6	
	Retirement village	7	7.9	7	3.1	
	Other	1	1.1	3	1.3	
Relationship						0.374
	Spouse	63	70.8	153	67.4	
	Parent	3	3.4	2	0.9	
	Adult Child	16	18.0	52	22.9	
	Sibling	3	3.4	5	2.2	
	Other	4	4.5	15	6.6	
Caring affected work						0.008
	Gave up job	16	18.0	41	18.1	
	Reduced hours	8	9.0	25	11.1	
	No change	2	2.2	21	9.3	
	Not working	60	67.4	112	49.6	
	Other	3	3.4	27	11.9	

Other caring responsibilities						0.373
	Yes	17	19.1	55	24.2	
	No	72	80.9	172	75.8	
Caring length (months)						0.544
	Mean (\pm SD)	18.4 \pm 24.39		21.3 \pm 43.64		
	Median (Range)	11.0 (1, 144)		10.0 (0.3, 420)		
PATIENT						
Patient gender						0.527
	Male	54	60.7	127	55.9	
	Female	35	39.3	100	44.1	
Patient age (years)						0.172
	Mean (\pm SD)	72.1 \pm 14.29		70.9 \pm 12.82		
	Median (Range)	74.0 (4, 93)		72.0 (35, 94)		
Diagnosis						0.039
	Cancer	66	74.2	171	75.3	
	Cancer + non-cancer	8	9.0	37	16.3	
	Non-cancer	15	16.9	19	8.4	
Length of diagnosis (months)						0.906
	Mean (\pm SD)	31.1 \pm 50.89		30.3 \pm 50.16		
	Median (Range)	13.0 (1, 400)		12.5 (0.3, 420)		
Length of palliative care (months)						0.001
	Mean (\pm SD)	6.0 \pm 8.29		3.0 \pm 4.28		
	Median (Range)	4.0 (0.3, 72)		1.5 (0.3, 29)		
Length of stay with Silver Chain (months)						<0.001
	Mean (\pm SD)	5.1 \pm 5.19		2.3 \pm 3.79		
	Median (Range)	3.2(0.03, 27.95)		0.9(0.03, 24.16)		

NB - Percentages may not equal 100% due to rounding.

Table 2. Summary of themes and corresponding sub-themes.

Theme	Sub-themes
Theme 1: Intrapersonal – Inward directed	<ul style="list-style-type: none"> -Prompting contemplation and reflection: “Making you think” -Gaining insight and awareness of support needs: Recognizing the “emotional and physical side of caring” -Feeling acknowledged and valued: : “Feeling as if you matter”
Theme 2: Connection with others – Outward directed	<ul style="list-style-type: none"> -Feeling less isolated: “Letting people in” -Improving support for future caregivers: “Making a difference for others” -Enhancing service improvement: “Needing documented evidence”
Theme 3: Interpersonal – Participant researcher relationship	<ul style="list-style-type: none"> -Expressing a “caring tone” -“Listening to my heart” -Respecting “my free will”

Figure 1

