

**Title: Planning and enabling meaningful patient and public involvement in dementia research**

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## **Abstract**

**Purpose of review:** To summarise the latest evidence on patient and public involvement (PPI) in dementia research. To identify methodologies used to deliver and evaluate co-production, potential barriers to involvement, and effective strategies to enable meaningful involvement of PPI representatives in dementia research.

**Recent findings:** PPI expertise in dementia research has been utilised to increase the acceptability of new interventions to end users and to interpret research findings from a PPI perspective. Practical adaptations to materials and meetings, and building relationships and trust were important facilitators for engagement. PPI has included caregivers, people living with dementia (PLWD) or both, with few studies involving the public or professionals. Engaging PLWD and hard-to-reach groups in PPI is challenging, with most current PPI representatives recruited from dementia organisations. Few studies have assessed the impact of PPI on study outcomes.

**Summary:** In this relatively new field, methods that allow meaningful engagement are being iteratively developed. Greater community and patient engagement will be a prerequisite for increasing diversity of PPI, to ensure voices of a broader range of stakeholders are heard. Documenting and publicising the impact of this work and engaging existing PPI representatives to support PPI recruitment could help with this.

**Keywords:** dementia, co-production, patient and public involvement

## **Introduction**

Patient and Public Involvement (PPI) in health and social care research involves the conduct of studies “with or by” rather than “to, about, or for” patients or members of the public (1). There has been a shift in discourse, from PPI in research, to one of “co-production” over the past ten years (2). Researchers have sought to move from passive consultation or collaboration, to active involvement of people who have an invested interest in and who can directly benefit from the work (3). The key principles of co-production in research include power sharing, inclusion of all relevant skills and perspectives, respecting different skills and experiences, and making joint decisions over roles and responsibilities (3). In dementia research, this might involve people living with dementia (PLWD), caregivers, health and/or social care professionals, voluntary sector workers and/or policy makers jointly sharing authority and responsibility for a project with the researchers, from the conception of ideas through to delivery and write up of findings and dissemination. Co-production in research can be challenging. It requires careful management of potentially opposing views between different stakeholders (4). Researchers need to ensure PPI representatives’ well-being, while avoiding over-protective and paternalistic attitudes that may hamper meaningful engagement. Participants living with dementia may require additional measures to facilitate participation where they experience difficulties with language, memory or other cognitive impairments, which are likely to worsen during long-term projects (5).

There has been only one previous review on PPI involvement in dementia research. A scoping review (that searched the literature up to February 2018) explored engagement in dementia research for PLWD and caregivers (6). Of 50 studies (almost all published since 2010), just under half involved caregivers and PLWD, 42% involved caregivers and 7% PLWD. Engagement varied, from membership on advisory committees, to collecting data as peer researchers. In three studies, PPI representatives conducted qualitative interviews, and were involved in the analytic process of making sense of findings. In some studies, PPI representatives co-authored publications. A limited number of studies

evaluated the impact of PPI involvement in research; those that did described how PPI influenced the development of accessible study materials, recruitment rates, study design decisions or interpretation of results.

Challenges to PPI included difficulties understanding the research process, lack of research training or experience and distress when exploring sensitive topics. Facilitators included early planning by researchers, adequate resources, early consideration of consent and capacity and involving dementia support organisations. Two studies reported that, despite intentions to engage PLWD and their best efforts to do so, they engaged only caregivers. The authors commented that there are compelling reasons to engage both PLWD and their caregivers, though involving both simultaneously may not allow either to fully and freely express themselves. They concluded that researchers should offer choice over whether and how to engage PLWD together with their caregivers.

Since publication of this scoping review, there has been a burgeoning interest in how to ensure the voice of people with lived experience of dementia is heard in dementia research. We therefore reviewed the literature to update the previous scoping review by identifying recent evidence on co-production in dementia research focusing on how PPI representatives are involved, how their views are elicited, potential barriers to involvement and strategies that enable effective co-production.

### **Literature search strategy**

We carried out a search for articles in Pubmed, Medline, Embase and Psycinfo on 21 April 2019. We restricted the search to articles published in English between 01 January 2018 to the search date. Search terms included: coproduction OR co-production OR PPI OR patient and public involvement OR lived experience OR peer research AND dementia OR Alzheimer\*.

### **Search results**

130 papers were identified of which 16 studies were retrieved for inclusion. The studies documented PPI involvement from the researcher's perspective (7-18), through analysis of workshop and advisory committee minutes (19, 20) or by directly asking PPI representatives for their experiences (21\*, 22\*\*). Half of the studies were co-authored by PPI representatives (8, 12\*, 14\*, 15\*, 19-22\*\*).

### **Who was involved?**

Three studies involved former or current caregivers of PLWD recruited via national dementia organisations (12\*), local services (19) or previous contact with researchers (17\*). Others involved mixed groups of PLWD and caregivers recruited through national or local dementia organisations or support groups (8, 9, 13, 15\*, 18, 21\*), or via contacts with researchers and local services (20). One study involved only PLWD recruited via national dementia organisations (16) and another involved health professionals and caregivers recruited via psychological services and national dementia organisations (11).

Only four studies involved a broader range of stakeholders including caregivers, the public, PLWD and carer support workers recruited via existing links with the research team, a national dementia organisation and a University volunteering service (22\*\*). One study involved members of the public, PLWD and caregivers, however it was unclear how they were approached (14\*), while another involved PLWD, their caregivers, older adults without dementia and healthcare professionals from local groups and services (10). One study ran groups with people affected by early onset dementia and their caregivers recruited through national dementia support groups, and also interviewed health professionals and voluntary sector workers (7).

### **How were they involved?**

PPI representatives were involved in research through membership on advisory committees, data analysis, developing and delivering interventions and influencing study design.

### ***Advisory roles***

Eleven studies involved caregivers and/or PLWD on advisory committees as “critical friends” to the study (7, 8, 12\*, 14\*-19, 21\*, 22\*\*). They reviewed funding (7, 8, 12\*, 18) ethics applications (8, 12\*, 22\*\*) or study protocols (7, 18, 22\*\*), provided feedback on recruitment strategies or materials (7, 8, 12\*, 19, 22\*\*), questionnaires (7, 21\*) or interview processes (16, 17\*, 21\*), designed study brands (22\*\*) and interpreted findings (7, 22\*\*).

### ***Analysing data***

While we did not identify further studies involving PPI representatives in data collection since the scoping review (6), two studies involved them in secondary analysis of interviews. In one study, an existing qualitative dataset on PLWD’s experiences of peer support networks was shared in workshops with four peer support groups (9) and in the other, four PLWD were involved in a two-hour analysis session of qualitative interviews on risk communication in dementia care (16). The session included role-play and interactive exercises alongside handouts of data extracts to illicit interpretations. Interpretations from the groups were described and built into the analytical process.

### ***Developing and delivering interventions***

Four studies described co-production of psychological interventions for caregivers of PLWD (10, 12\*, 14\*, 15\*). In one study, health professionals and caregivers were asked for feedback during development of materials, testing and piloting of a computerised cognitive behavioural therapy package (12\*), while another involved caregivers and PLWD in the development of an intervention to reduce sleep problems in PLWD (15\*). One caregiver was part of the intervention development team and feedback on intervention content was gathered via focus groups or email. Another study involved both PLWD and their caregivers in co-designing and evaluating feasibility of a psychoeducational toolkit on dementia support (10), while one study piloted a community-based approach to tackle negative attitudes towards dementia and improve knowledge of the condition

(14\*). In this study, PLWD and caregivers were involved in creating and delivering educational sessions, participating in media campaigns, and developing and distributing materials.

### ***Influencing experimental study design***

Two studies involved PLWD and caregivers in study design, with one using experiences from a support group for people with dementia and balance problems to develop and adapt sensorimotor equipment for use within this population (8), while the other consulted PLWD and caregivers in the design of an accessible Delphi survey on outcome measures for studies of non-pharmacological interventions (13). Modifications included translation of outcomes into accessible statements, development of simplified rating scales, and preferences on how to present findings.

### **Barriers to co-production**

Ten studies described challenges to co-production, mostly from the researcher (7, 9, 10, 12\*, 15\*-17\*) rather than caregiver/PLWD perspectives (19, 21\*, 22\*\*).

Some authors discussed hypothetical barriers to participation, including concerns that PPI representatives may struggle with roles and capabilities, feel unable to speak up in meetings or be uncertain how to proceed when they held different views to the researchers (12\*, 19). They may become experts over time and be less representative of those whose views they originally sought to represent (12\*).

Engaging PLWD and hard to reach groups was identified as a challenge including a lack of engagement with people with severe dementia (9, 16), difficulties recruiting any PLWD to an advisory committee, and challenges in obtaining feedback from PLWD due to difficulties understanding and remembering survey questions (10). Two studies acknowledged that PPI representatives were recruited from existing dementia research groups, committed to research and possibly not representative of the wider community (7, 15\*). One study described a particular challenge of engaging with the deaf

community (17\*), while another reported difficulties engaging health professionals and older adults without dementia who queried how the project was relevant to them (10).

One study gave “practical reasons” for why PLWD who wanted to be involved in analysing and interpreting results were not given this opportunity (18). Where PLWD were engaged in data analysis, they were not provided with training or, due to time, resources and perceived burden, not involved in selecting extracts for analysis (16).

Caregivers involved in longitudinal research experienced a loss of connection to the study during “quiet times” and frustratingly long waits for results (21\*), Practical and financial arrangements were identified as the biggest challenge in another study (22\*\*), including untimely reimbursement and difficulties accessing training resources. To satisfy a diverse range of experiences, the PPI group (n=12) in this study was large, resulting in difficulties managing expectations and relationships, and planning dates that everyone could attend. Cultural barriers were described with a lack of funds for interpreters meaning exclusion of non-English speakers.

One study acknowledged the precarious balance of power between clinical academics and PPI representatives and described carefully navigating contradictory views, while remaining transparent about the decision-making process (15\*). Practical challenges to attending meetings were also identified if caregivers or relatives became unwell. Another study reported that PPI representatives gave no negative feedback (16) and acknowledged that this was due to a lack of systematic evaluation of the PPI process.

### **Effective co-production**

Eleven studies described strategies that had enabled PPI involvement in research (7, 9, 12\*, 13, 15\*-19, 21\*, 22\*\*).

Practical strategies included adapting reading materials (e.g. large font (9, 21\*), black writing on coloured paper (9), pictures, word cards or story boards (9, 21\*), use of role play (16), meeting in

accessible and familiar environments (18, 21\*), and reimbursing for time and travel (15\*, 21\*, 22\*\*). Strategies for effective meetings included clear agendas and terms of reference (12\*, 19), allowing extra time (13, 21\*) and a sensitive facilitator to encourage comment and reassure members that technical expertise was not required (12\*, 18, 21\*). Flexibility on how people could be involved (e.g. face-to-face, telephone or email) (15\*, 21\*) and contact between meetings via email or phone (13, 21\*) were identified as important for maintaining engagement.

One study identified the skills of each group member so that they could choose specific tasks and so that support could be provided to members who wanted to participate in areas that they lacked confidence (e.g. presenting at conferences) (22\*\*). Another study highlighted the importance of involving “community insiders” as researchers to engage particularly hard to reach groups; recruitment of caregivers of deaf people with dementia to a PPI group was coordinated by a deaf researcher who was fluent in British Sign Language (17\*).

The importance of good communication and building relationships and trust between researchers and PPI representatives included creating an informal environment (21\*), discussing differences in opinion and providing feedback on how PLWD and caregivers had contributed to, and influenced the research (7, 13, 15\*, 19, 21\*). One study recognised that PLWD may have additional communication needs and that researchers need to be trained to accommodate these needs (18).

### **Impact and dissemination**

Three studies described caregivers and PLWD deriving personal benefit from involvement in research; of making a contribution, being given purpose and having a voice (10, 19, 21\*). The experience had also enabled them to provide support to other caregivers (19).

Six studies described the impact of PPI involvement on the research process. This included the use of personal experiences to tailor research materials (21\*), interview questions and techniques (16) and intervention manuals for enhanced implementation and future testing (10, 11). The most tangible

impact was on increasing recruitment rates. PPI representatives directly recruited from hard to reach groups (22\*\*), with one study reporting that 69% of enquiries from potential participants were generated through PPI representatives promoting the study to community organisations (12\*). Amendments to study procedures following feedback from PPI representatives resulted in lower attrition rates (12\*).

Seven studies described the wider impact of involving PLWD and caregivers in research (8, 10, 12\*, 14\*, 17\*, 21\*, 22\*\*). Caregivers co-presented at conferences or training events (12\*, 14\*, 17\*, 21\*, 22\*\*), disseminated findings through local media (14\*, 19, 22\*\*), campaigned and raised funds for further research (19) and researchers reported building up their PPI networks for future studies (10, 22\*\*). Two projects created films to document PPI representative experiences of being involved in research (8, 21\*) and to educate the public on dementia. It was unclear how these films were disseminated. Only one study that co-produced education and awareness raising events assessed impact through questionnaires administered to community members before and after the events (14\*). There was increased awareness of the experiences of PLWD and dementia services, and those who attended an event reported less negative views about dementia than those who did not.

## **Conclusion**

In this relatively new field, methods that allow meaningful PPI engagement in research are being iteratively developed. Recent studies are using PPI experiences to co-produce interventions and influence study design and interpretation of findings. One impact of PPI noted in included studies was enhanced recruitment, especially in hard to reach groups. By contrast, PPI representation, often recruited from advocacy groups and research organisations has been criticised for not representing the wider community of PLWD (23, 24), though recent studies have successfully recruited from local services (10, 11, 19, 20). Greater community and patient engagement with PPI will be a prerequisite for more representative PPI. Documenting and publicising the impact of this work is likely to help with this.

An absence of involvement from non-academic health and social care professionals, voluntary sector workers and the public has also been noted, with most PPI involvement reflecting patient (and caregiver) rather than public involvement. Most studies did not consider adaptations to enable involvement of people with advanced dementia. Alternative methods of communicating research through vignettes and photo elicitation could be considered (25).

The researcher perspective was the dominant voice within publications, rather than the voice of PPI representatives; however, they often appeared as co-authors. Very little reflection occurred around the co-production process, with only one study recognising the difficulties of power sharing (15\*) and another identifying the skills and training needs of the group (22\*\*). Future work should consider using established guidelines to help effectively design and evaluate PPI activities within research (26).

The wider impact of PPI involvement is still poorly described (27). Future research needs to involve, evaluate and document the experiences of caregivers, PLWD, professionals and the public. We need to hear their voices, rather than just the researcher's perspective of PPI in research.

### **Key points**

- Patient and public involvement in dementia research is evolving, with recent reports describing PPI involvement of co-producing new interventions and interpreting research findings.
- Most of the research involves caregivers of people living with dementia, however methods and resources to improve engagement of people living with dementia, hard to reach groups, health and social care professionals and the public as co-producers in dementia research are needed.
- Practical adaptations to materials and meetings, building relationships and trust and offering flexibility can enhance engagement of people living with dementia and family carers in research.
- Future work should directly elicit the experiences of PPI representatives in the research process and formally evaluate the strengths and limitations of PPI from all relevant stakeholder perspectives.

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**Conflicts of interest**

AB, MO and CC are involved in the co-production of an intervention to support caregivers and people living with dementia to stay independent at home for longer: NIDUS (New Interventions in Dementia Study)

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Researchers measured the impact of dementia awareness and training events on a local community. PPI representatives were involved in both the design and delivery of the events and dissemination of findings.

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One of only two studies that explored PPI involvement by directly asking PPI representatives for their views on their experiences of being involved in the research project. Strategies and barriers to effective PPI involvement as well as the impact of involvement on the research process and dissemination of findings were also documented.

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The researchers involved a broad range of stakeholders in a diverse range of research activities, and explored PPI involvement by directly asking PPI representatives about their experiences. Strategies and barriers to effective PPI involvement, as well as the impact of involvement on the research process and dissemination activities were documented. The skills of PPI representatives were taken into consideration when planning involvement activities.

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