Patient and Public Involvement in Identifying Dementia Research Priorities

Jennifer Bethell, PhD,* Dorothy Pringle, PhD,† Larry W. Chambers, PhD,** Carole Cohen, MD,††† Elana Commissio, MA,* Katherine Cowan, MA,** Phyllis Fehr, RN,¶¶¶ Andreas Laupacis, MD,†††††† Paula Szeto, OT Reg (Ont),* and Katherine S. McGilton, PhD†††

OBJECTIVES: To engage persons with dementia, friends, family, caregivers, and health and social care providers to identify and prioritize their questions for research related to living with dementia and prevention, diagnosis, and treatment of dementia.

DESIGN: The Canadian Dementia Priority Setting Partnership (PSP) followed James Lind Alliance PSP methods. Results were compared with the World Health Organization research prioritization exercise and the United Kingdom Dementia PSP.

SETTING: Canada.

PARTICIPANTS: In the first survey, 1,217 individuals and groups from across Canada submitted their questions about dementia. 249 participated in the interim prioritization. For the final prioritization workshop, the 28 participants included persons with dementia, friends, family, caregivers, health and social care providers, Alzheimer Society representatives, and members of an organization representing long-term care home residents.

RESULTS: The Canadian Dementia PSP top 10 priorities relate to health, quality of life, societal issues, and dementia care. Five priorities overlap with one or both of the other two prioritization initiatives.

CONCLUSION: These results provide researchers and research funding agencies with topics that individuals with personal or professional experience of dementia prioritize, but they are not intended to preclude research into other aspects of dementia. J Am Geriatr Soc 66:1608–1612, 2018.

Key words: dementia; research; patient engagement; patient and public involvement; research priorities

Patient and public involvement in health research is being advocated for on methodological and moral grounds, including by national funding agencies in the United States (Patient-Centered Outcomes Research Institute (PCORI))², Canada (Canadian Institutes of Health Research, Strategy for Patient-Oriented Research),³ and the United Kingdom (National Institute for Health Research (NIHR), INVOLVE http://www.involve.org.uk/about-involve/),⁴ yet there is still a gap between current clinical research and the priorities of patients, caregivers, and clinicians.⁵,⁶

The Canadian Dementia Priority Setting Partnership (PSP) was established to engage individuals with personal or professional experience of, or interest in, dementia—including persons with dementia, friends, family, caregivers and health and social care providers—and to use the James Lind Alliance (JLA) methods to identify and prioritize questions for research on dementia that are important to these individuals and informed by their experiences. Accordingly, the scope of the Canadian Dementia PSP included questions related to living with dementia, as well as prevention, diagnosis, and treatment.
In this article, to draw on others’ perspectives, we compare the results of the Canadian Dementia PSP with 2 other recent dementia research prioritization processes: the World Health Organization (WHO) research prioritization exercise\(^7\) and the JLA UK Dementia PSP.\(^8\) The WHO research prioritization exercise used an adapted version of the Child Health and Nutrition Research Initiative (CHNRI) method and consulted international researchers and stakeholders (but did not explicitly include persons with dementia or their friends, family, and caregivers), whereas the UK Dementia PSP used methods similar to those used in the current study in a UK population. Although there are other published dementia research agenda recommendations,\(^9,10\) the scope of the 2 included here were most comparable with the Canadian Dementia PSP. Both the WHO and UK exercises included prevention, diagnosis, treatment and care of dementia. The WHO included basic science research (mechanisms or causes of dementia) within these themes, whereas the UK Dementia PSP, like the Canadian Dementia PSP, did not.

**METHODS**

The Canadian Dementia PSP followed the methods of the JLA, a nonprofit initiative supported by the NIHR (UK) that has facilitated nearly 60 PSPs involving patients, caregivers, and health and social care providers in developing shared research priorities.\(^11\) The JLA methods (http://www.jla.nihr.ac.uk/jla-guidebook/) have been described, in a report to PCORI, as a model for collaboration between the public, clinicians, and researchers in setting research priorities.\(^12\)

A steering group oversaw and advised the study, and partner organizations were involved in several aspects of the study, including promoting surveys and nominating workshop participants (Supplementary Table S1). Persons with dementia were also involved in this process. One person with dementia was a member of the steering group, and one of the partner organizations, the Ontario Dementia Advisory Group (http://www.odag.ca), facilitated teleconference and videoconference discussions with persons with dementia to review and provide advice on the content and format of the surveys. Research ethics board approval was granted through the University Health Network, Toronto, Canada.

A survey was conducted online (https://www.limesurvey.org/) and on paper, in English and French, from May to August 2016 and promoted by partner organizations through social media, mailed surveys, and group discussions. Respondents were asked for questions about living with dementia, as well as dementia prevention, diagnosis, treatment, and to provide demographic information and contact details (if interested in participating in the interim prioritization process). Two research team members working together categorized and summarized the submission data. Submissions that were out of scope or could not be formulated into a question and questions that were asked only once were excluded. The resulting list of questions was then checked against available research evidence, and a list of 79 questions about dementia was created (Supplementary Appendix S1, Supplementary Table S2).

For the interim prioritization, a survey was conducted online (https://www.limesurvey.org/) and on paper, in English and French, from April to May 2017. Respondents were asked to identify, from the list of 79 questions, their unranked top 10 priorities. The number of times each question was selected was tabulated, and a ranked list of the questions was created. The demographic information from this phase showed that very few persons with dementia had participated, so additional input from persons with dementia was sought. Specifically, a support group of persons with dementia, held by a provincial Alzheimer society, discussed the questions and themes. Although most of their priorities were also highly ranked in the overall list, 3 additional questions that were important to the group were added to the short list. In total, 23 questions were identified for discussion at the final prioritization workshop (June 8–9, 2017, Toronto, Canada). Adaptations were made to the final prioritization workshop to facilitate involvement of persons with dementia.\(^13,14\)

Results of the Canadian and UK Dementia PSPs were reported as top 10 lists for research so the comparison was straightforward. However, because of the expanded scope of the WHO research prioritization exercise (including basic science research) and the presentation of the priorities in the main paper (top 3 ranked research avenues in each of 7 overarching research domains), this comparison was less straightforward. Therefore, to create a comparable top 10 list from the WHO research prioritization exercise, 2 of its research domains were excluded (pharmacological and nonpharmacological clinical-translational research, physiology and progression of normal aging and disease pathogenesis), and to limit the number of priorities to 10 but also reflect their presentation of the results, the top 2 ranked priorities within each of the 5 remaining research domains were retained.

**RESULTS**

In the first survey, 1,217 individuals and groups from across Canada submitted their questions about dementia and, in the interim prioritization, 249 took part (Supplementary Table S3, Supplementary Figure S1). A group of persons with dementia (n=5); friends, family, and caregivers (n=5); health and social care providers (n=9); Alzheimer Society representatives (n=5); and members of an organization representing long-term care home residents (n=2) decided on the top 10 priorities for dementia research at the final prioritization workshop (Table 1). Five of the top 10 priorities from the Canadian Dementia PSP overlapped with 1 or both of the WHO research prioritization exercise and UK Dementia PSP (Table 2). All 3 prioritization exercises identified the importance of assisting caregivers of persons with dementia and addressing behavioral and psychological symptoms of dementia.

**DISCUSSION**

The Canadian Dementia PSP identified priorities for dementia research related to the health and quality of life of...
Table 1. Top 10 Priorities for Research from the Canadian Dementia Priority Setting Partnership

1. What is the impact of stigmas associated with dementia and mental health issues on persons with dementia and their families? What are effective ways of reducing the stigma experienced by persons with dementia and their friends, family caregivers/care partners?
2. What can be done to support emotional well-being, including maintaining a sense of dignity, for persons with dementia?
3. Among persons with dementia, what is the impact of early treatment on quality of life, disease progression and cognitive symptoms?
4. How can the health system build and sustain the capacity to meet the health and social care needs of persons with dementia and their friends or family caregivers/care partners?
5. What services, supports and therapies for friend or family caregivers/care partners of persons with dementia would improve or maintain health, well-being and quality of life for persons with dementia and their friends or family caregivers/care partners?
6. After dementia is diagnosed, what would help persons with dementia and their friends, family and caregivers/care partners get the information, treatment, care and services they may need?
7. What dementia-related skills and knowledge should health and social care providers have? What are effective ways of providing them with these skills and this knowledge? How can the number of health and social care providers who have these skills and this knowledge be increased?
8. What enables the creation of dementia-friendly communities? What impact do dementia-friendly initiatives have on persons with dementia and their friends, families and caregivers/care partners?
9. What would ensure implementation and sustainability of best practices for dementia care within and across health care settings, including effective approaches to providing person-centered care?
10. Among persons with dementia, what are the effects of non-pharmacological treatments compared to pharmacological treatments on behavioural and psychological symptoms of dementia? Can non-pharmacological treatments replace, reduce or be used in conjunction with pharmacological treatments for managing behavioural and psychological symptoms of dementia?

Table 2. Overlap of Research Priorities Identified from Canadian Dementia Priority Setting Partnership (PSP), UK Dementia PSP, and World Health Organization (WHO) Research Prioritization Exercise

| Research Priorities | Canadian PSP | UK PSP | WHO |
|---------------------|--------------|--------|------|
| **Complete overlap (identified in all three prioritization exercises)** | | | |
| Caregivers of persons with dementia: determine most effective service, support, therapy, education, and training interventions | ● | ● | ● |
| Behavioral and psychological symptoms of dementia: determine and ensure optimal use and combinations of pharmacological and nonpharmacological approaches | ● | ● | ● |
| **Partial overlap (identified in 2 of 3 prioritization exercises)** | | | |
| Dementia-friendly communities: determine effectiveness, cost-effectiveness, and factors that enable their creation | ● | ○ | ● |
| Stigma and discrimination associated with dementia: assess effect and effective strategies to target and reduce | ● | ○ | ● |
| Best practices for dementia care: determine best approaches for ensuring dissemination, implementation, and sustainability | ● | ● | ○ |
| Dementia diagnosis in primary care: identify clinical practice and health system-based interventions to promote effective, timely, accurate diagnosis and assess effect of early diagnosis | ○ | ● | ● |
| Care for persons with advanced dementia at end of life: determine best approaches to care and identify strategies to anticipate and deliver most effective and cost-effective care | ○ | ● | ● |
| **No overlap (identified in 1 prioritization exercise only)** | | | |
| Ways to support emotional well-being of persons with dementia | ● | ○ | ○ |
| Impact of early treatment on quality of life, disease progression, and cognitive symptoms | ● | ○ | ○ |
| Ways to build and sustain health system capacity to meet health and social care needs of persons with dementia and their caregivers | ● | ○ | ○ |
| Ways to ensure information, treatment, care, and services are provided to persons with dementia and their friends, family and caregivers after dementia is diagnosed dementia-related skills and knowledge among health and social care providers | ● | ○ | ○ |
| Most effective components of care to maintain independence among persons with dementia | ○ | ● | ○ |
| Best way to care for persons with dementia in a hospital setting when they have acute healthcare needs | ○ | ● | ○ |
| Most effective ways to encourage persons with dementia to eat, drink, and maintain nutritional intake | ○ | ● | ○ |
| Optimal time for persons with dementia to move into a care home and how to improve standard of care | ○ | ● | ○ |
persons with dementia and their friends, family, and caregivers; societal issues; and delivery and quality of dementia care. There is substantial overlap between the Canadian Dementia PSP priorities and the WHO research prioritization exercise and the UK Dementia PSP, although there are 5 priorities unique to the Canadian Dementia PSP; potential explanations for this include that there are uniquely Canadian priorities (e.g., concern about the sustainability of the publicly funded healthcare system)\(^1\) or methodological differences between the 3 exercises (the processes themselves, the questions they elicited, the people and groups they involved).\(^2\) The Canadian Dementia PSP, the UK Dementia PSP, and the WHO research prioritization exercise each included questions related to prevention, but ultimately only the WHO research prioritization exercise identified prevention among the research priorities.

The Canadian Dementia PSP used established methods to engage Canadians in setting priorities for research on dementia, but the process had limitations. Those who participated may not have been representative of all those the PSP was meant to engage. In particular, the cognitive and communication difficulties associated with dementia would have precluded some persons with dementia from taking part, especially those with late-stage dementia. We endeavored to enable participation by engaging persons with dementia in developing our surveys, encouraging group responses through partner organizations (e.g., support groups for persons with dementia), seeking additional input for the interim prioritization, implementing recommendations for a dementia-friendly workshop,\(^3\)\(^4\) and ensuring representation of persons with dementia at the workshop. Still, there is limited evidence on the best approaches for engaging persons with dementia in research.

The UK Dementia PSP was the first research prioritization to involve persons with dementia, friends, family, caregivers, health and social care providers, and the public and to use the JLA methods in this context.\(^5\) The WHO research prioritization exercise adapted CHNRI methods to involve international experts in a global research prioritization process.\(^6\) Like the Canadian Dementia PSP, these prioritization processes elicited hundreds of questions about dementia from hundreds of experts. The priorities identified here, particularly taken in context with the other dementia research prioritization exercises, offer directions for researchers and research funding agencies, although they do not obviate the need for continued research into other areas, including mechanisms of disease and new treatments, which are unequivocally important.\(^7\) Finally, more research is needed to develop strategies and methods for involving persons with dementia and their friends, family, and caregivers in research and making research dementia-friendly.\(^8\)

**ACKNOWLEDGEMENTS**

We thank the steering group members and partner organizations for their many contributions, in particular the Ontario Dementia Advisory Group for their input and guidance in creating the surveys. We also express gratitude to the many Canadians who took part in the Canadian Dementia PSP, in particular those who took part in the final workshop. We also thank Jessica Babineau, Information Specialist, Toronto Rehabilitation Institute, for her assistance with the literature search.

**Financial Disclosure**: This work was supported by the Alzheimer Society Research Program (ASRP) as part of the Alzheimer Society of Canada’s commitment to the Canadian Consortium on Neurodegeneration in Aging (CCNA). Jennifer Bethell was funded by an ASRP/CCNA postdoctoral award. Katherine McGilton is supported by the Walter and Maria Schroeder Institute for Brain Innovation & Recovery.

Methods and preliminary results were presented at the Alzheimer’s Association International Conference (London, UK, July 2017) and Canadian Conference on Dementia (Toronto, Canada, November 2017).

**Conflict of Interest**: None.

**Author Contributions**: all authors: study concept and design, acquisition of subjects and data, analysis and interpretation of data, and preparation of manuscript.

**Sponsor’s Role**: The Alzheimer Society participated in study oversight through membership on the Steering group and as a partner organization.
REFERENCES

1. Wilson P, Mathie E, Keenan J et al. ReseArch with Patient and Public involvement: a RealisT evaluation—the RAPPORT study. London, UK: NHS National Institute for Health Research; 2015.

2. Forsythe LP, Ellis LE, Edmundson L et al. Patient and stakeholder engagement in Canada: A scoping review of the ‘how’ and ‘what’ of patient engagement in health research. Health Res Policy Syst 2018;16:5.

3. Manafo E, Petermann L, Mason-Lai P, Vandall-Walker V. Patient engagement in Canada: A scoping review of the ‘how’ and ‘what’ of patient engagement in health research. Health Res Policy Syst 2018;16:5.

4. INVOLVE. National Institute for Health Research (online). Available at http://www.involve.org.uk/frequently-asked-questions/ Accessed November 28, 2017.

5. Jun M, Manns B, Laupacis A et al. Assessing the extent to which current clinical research is consistent with patient priorities: A scoping review using a case study in patients on or nearing dialysis. Can J Kidney Health Dis 2015;2:70.

6. Crowe S, Fenton M, Hall M, Cowan K, Chalmers I. Patients’, clinicians’ and the research communities’ priorities for treatment research: There is an important mismatch. Res Involv Engagem 2015;1:2.

7. Shah H, Albanese E, Duggan C et al. Research priorities to reduce the global burden of dementia by 2025. Lancet Neurol 2016;15:1285–1294.

8. Kelly S, Lafortune L, Hart N, Cowan K, Fenton M, Brayne C. Dementia priority setting partnership with the James Lind Alliance: Using patient and public involvement and the evidence base to inform the research agenda. Age Ageing 2015;44:985–993.

9. Feldman HH, Haas M, Gandy S et al. Alzheimer’s disease research and development: A call for a new research roadmap. Ann N Y Acad Sci 2014;1313:1–16.

10. Workgroup on NAPA’s scientific agenda for a national initiative on Alzheimer’s disease. Alzheimers Dement 2012;8:357–371.

11. Cowan K. The James Lind Alliance: Tackling Treatment Uncertainties Together. J Ambul Care Manage 2010;33:241–8.

12. Nass P, Levine S, Yancy C. Methods for Involving Patients in Topic Generation for Patient-Centered Comparative Effectiveness Research—An International Perspective. Research Priorities White Paper (PCORI-SOL-RPWG-001) for the Patient-Centered Outcomes Research Institute (PCORI) 2012 (online). Available at https://www.pcori.org/assets/Methods-for-Involving-Patients-in-Topic-Generation-for-Patient-Centered-Comparative-Effectiveness-Research-%E2%80%93-An-International-Perspective.pdf Accessed April 18, 2018.

13. Dementia Friendly Meetings Guidelines. The Scottish Dementia Working Group (online). Available at http://www.sdwg.org.uk/wp-content/uploads/2016/04/Dementia-Friendly-Meetings.pdf Accessed November 16, 2017.

14. Adapting PSP surveys and workshops to make sure everyone can get involved. James Lind Alliance (online). Available at http://www.jla.nihr.ac.uk/news/adapting-psp-surveys-and-workshops-to-make-sure-everyone-can-get-involved/6635 Accessed November 16, 2017.

15. Soroka SN. Public Perceptions and Media Coverage of the Canadian Healthcare System: A Synthesis. Canadian Health Services Research Foundation 2011 (online). Available at http://www.cfhi-fcass.ca/SearchResults-News/11-10-06/da81c69-c4ca-4686-aa4f-02b961401c71.aspx Accessed November 16, 2017.

16. Elliott MJ, Straus SE, Pannu N et al. A randomized controlled trial comparing in-person and wiki-inspired nominal group techniques for engaging stakeholders in chronic kidney disease research prioritization. BMC Med Inform Decis Mak 2016;16:113.

17. Liggins C, Snyder HM, Silverberg N et al. International Alzheimer’s Disease Research Portfolio (IADRP) aims to capture global Alzheimer’s disease research funding. Alzheimers Dement 2014;10:405–408.

18. Scottish Dementia Working Group Research Sub-Group UK. Core principles for involving people with dementia in research: Innovative practice. Dementia 2014;13:680–685.

SUPPORTING INFORMATION

Table S1. Steering Group membership and list of Partner Organisations
Appendix S1. Literature search strategy
Table S2. Long list of questions
Table S3. Description of survey and interim survey respondents
Figure S1. Canadian Dementia Priority Setting Partnership Infographic

Please note: Wiley-Blackwell is not responsible for the content, accuracy, errors, or functionality of any supporting materials supplied by the authors. Any queries (other than missing material) should be directed to the corresponding author for the article.