

UC San Diego

UC San Diego Previously Published Works

Title

Positive psychiatry comes of age

Permalink

<https://escholarship.org/uc/item/5xq6t47c>

Journal

International Psychogeriatrics, 30(12)

ISSN

1041-6102

Author

Jeste, Dilip V

Publication Date

2018-12-01

DOI

10.1017/s1041610218002211

Peer reviewed

GUEST EDITORIAL

Positive psychiatry comes of age

Positive psychiatry is the science and practice of psychiatry that focuses on psycho-bio-social study and promotion of well-being and health through enhancement of positive psychosocial factors (such as resilience, optimism, wisdom, and social support) in people with illnesses or disabilities as well as the in community at large (Jeste and Palmer, 2015). It is based on the principles that there is no health without mental health and that mental health can be improved through preventive, therapeutic, and rehabilitative interventions to augment positive psychosocial factors. Positive psychiatry is not a naïve, feel-good pseudoscience that views the world through rose-colored glasses. It is an evidence-based approach to understanding normal behavior as well as psychopathology and to improving well-being by measuring and enhancing positive psychosocial factors (Jeste *et al.*, 2015).

Today most of the articles in most of the geriatric psychiatry journals are focused on neuropsychiatric disorders and disabilities in later life. This is not surprising as these conditions are indeed associated with clinically significant functional impairments and are a major cause of adverse mental, physical, social, and financial outcomes for the patients and their families. Research on and treatment of these maladies should, therefore, be a priority for our field. However, a near exclusive focus on disorders is not just unhelpful but also counter-productive for psychiatry in general and geriatric psychiatry in particular.

With rapidly growing numbers of older people across the globe, healthcare systems that are dependent on individual-level treatment of diseases are painfully inefficient and unsustainable. There is now a growing consensus that prevention is the key to revolutionizing healthcare. This has led to a discussion about promoting healthy lifestyle, such as physical activity, calorie restriction, and stopping smoking and substance use. Changing the lifestyle involves changing a person's behavior. Who are the experts in interventions to modify behavior? The answer is mental health practitioners. We treat the most serious behavior problems such as delusions, hallucinations, and suicidal behaviors. We have at least reasonable knowhow about biological and psychotherapeutic tools for helping modify unhealthy behaviors.

As important as healthy lifestyle but rarely attended to in the arena of preventive healthcare is the role of positive personality traits such as resilience, optimism, compassion, self-efficacy, and wisdom as well as social and environmental support. There is strong empirical evidence for the association of these positive factors with better mental and physical health, cognitive function, and even longevity. Who are the experts in assessing and enhancing these positive psychosocial factors? Once again, it is mental health experts. Thus, positive psychiatry should be at the center of the new healthcare system.

This issue of *International Psychogeriatrics* is the first ever issue of our journal with the theme of positive psychiatry. The current issue contains three data-based research articles (Bailly *et al.*, 2018; Ihle *et al.*, 2018; Montross-Thomas *et al.*, 2018) from France, USA, and Switzerland, respectively. These papers report stability of spirituality in older adults followed over a five-year period (Bailly *et al.*, 2018), response of hospice patients to the diagnosis of terminal illness by cultivating wisdom through a balance between active acceptance of the current situation and continued push for a galvanized growth (Montross-Thomas *et al.*, 2018), and significant contributions of close friends and leisure activity engagement to better cognitive performance in old age (Ihle *et al.*, 2018). There are accompanying commentaries (Baiyewu, 2018; Forlenza and Vallada, 2018; Pachana and Mitchell, 2018) from Brazil, Australia, and Nigeria, respectively, which discuss both limitations and implications of those studies.

In the future, as we continue publishing many papers on neuropsychiatric diseases of aging, we will also include, from time to time, articles on topics related to positive psychiatry. We hope to have papers and commentaries from different parts of the world with articles on various positive topics including successful aging, mind-body interventions, and age-friendly communities, among others. I welcome input from our readers.

Thanks to pioneers like Seligman (Seligman and Csikszentmihalyi, 2000), positive psychology is now well accepted even by lay public. Yet, there are few papers on positive constructs such as optimism, resilience, and wisdom in psychiatric journals

and few chapters on these topics in psychiatric textbooks. The goal of positive psychiatry is to enrich psychiatric literature and practice by incorporating positive psychosocial factors in the study and treatment of people with and without mental illnesses. Moreover, being a branch of medicine, positive psychiatry will emphasize health as well as biology along with psychology, sociology, and (in the case of psychogeriatrics) gerontology.

Positive psychiatry is not a geographically localized phenomenon but a global movement. There is now a formal section on Positive Psychiatry in the World Psychiatric Association and a Caucus on Positive Psychiatry in the American Psychiatric Association. Other national organizations are beginning to get involved too. During the last few years, a number of symposia in this area have been presented at different national and international conferences. At least two books have been published (Jeste and Palmer, 2015; Summers and Jeste, 2018) and several others are in the works.

In terms of measurements, we already have access to a number of reliable and validated self-report inventories for various positive factors (Eglist *et al.*, 2018). Self-report measures have been criticized for having both conscious (e.g. deliberate deception) and unconscious (e.g. impression management) biases in human introspection and subsequent reporting. However, research has shown a significant association between subjective and objective measures of constructs such as well-being. Self-report inventories for internal states such as happiness and subjective recovery are inherently tied to an individual's introspective feelings rather than to an external biological proxy, at least at the present time. For example, most of us will agree that the best way to determine individuals' level of happiness is by asking them about their current inner experiences and feelings rather than by measuring cerebrospinal fluid levels of catecholamines (although these may correlate with subjective happiness).

Positive psychiatry applies even more importantly to people with serious mental illnesses such as schizophrenia and major depression, serious physical illnesses such as cancer and HIV-AIDS, and serious cognitive disorders including dementias or major neurocognitive disorders (Palmer *et al.*, 2014; Cohen *et al.*, 2017; Sharma *et al.*, 2017; Moore *et al.*, 2018). In one study, Mohr and colleagues (2011) measured religiosity and spirituality in 115 outpatients with schizophrenia and related disorders, using semi-structured clinical interviews, and then followed the cohort for three years. The investigators found that participants who engaged

in healthy religious coping strategies and who valued spirituality experienced less severe negative symptoms and better interpersonal functioning and quality of life than other patients.

In addition to their effects on emotional and cognitive health, positive psychosocial factors are closely related to physical health and specific biomarkers of health as well (Edmonds *et al.*, 2018). Empirical evidence supports links between biomarkers and measures of positive psychiatry, including allostatic load, telomere length, inflammation, and genes (Schutte *et al.*, 2016; Wiley *et al.*, 2017), although such research in people with serious mental illnesses has so far been sparse. One study reported that self-efficacy moderated the relationship between subjective stress and interleukin-6 levels among dementia caregivers (Mausbach *et al.*, 2007). In a two-year randomized controlled trial in adults with chronic schizophrenia, Eack *et al.* (2010) showed that, compared to a control group, a multimodal intervention approach – cognitive enhancement therapy – not only improved cognitive performance but also seemed to protect against gray matter loss on MRI.

Chronological aging is associated with increases in both physical and cognitive impairments. However, a large number of older adults report “successful aging” (Jeste *et al.*, 2013). This concept was originally defined as absence of significant disease burden, but – consistent with the positive psychiatry movement – more recent data suggest that successful aging does not require absence of illness, but rather the overall positive psychological outlook of the individual. There is a frequently observed paradox of aging – as physical health declines, mental well-being improves. The importance of emotional health in aging is illustrated by the “positivity effect” – i.e., a tendency for older adults to experience a higher ratio of positive to negative emotions relative to younger adults (Mather and Carstensen, 2005). While there is considerable research on the so-called longevity genes, positive personality traits including resilience, optimism, and wisdom may also be of relevance for exceptional longevity (Scelzo *et al.*, 2018).

There is also a wealth of neurobiological data showing that, contrary to traditional beliefs, neuroplasticity continues into old age (albeit to a lesser degree than in youth, enabling new learning and adaptation in the context of appropriate environmental stimulation. Mechanisms underlying brain plasticity in older adults include neural compensation for age-related decline through recruitment of additional brain circuits in the performance of tasks, increased dendritic

arborization, synaptic proliferation, greater vascularity, and formation of new neurons in specific regions – e.g., hippocampal dentate gyrus (Gage, 2002).

There is an urgent need for promoting research in positive psychiatry. Likewise, on the clinical side, while clinicians will generally need to ask standard questions about symptoms and functional impairments, it is imperative that they also undertake a deliberate assessment of positive psychosocial factors. To translate the experimental results to a clinical setting will require development of a training and administrative infrastructure to support such assessments and interventions in positive psychiatry.

An important caveat to the theory and practice of positive psychiatry is the realization that the relationship between positive factors and overall functioning/well-being may exist in an inverted U shape. Enhancing these characteristics leads to functional improvements up to a certain point, beyond which increases can have adverse consequences. For example, excessive optimism can precipitate risky medical and health-related choices, ultimately leading to negative outcomes. Similarly, extreme happiness may be associated with a lack of attention to prophylactic healthful behaviors, leading to increased morbidity and even mortality. Excess of certain positive factors is intuitively related to the symptom profiles of psychiatric disorders such as bipolar disorder and narcissistic personality disorder. However, most individuals with serious mental illnesses are far more likely to have lower rather than higher levels of positive factors including optimism, resilience, happiness, personal mastery, coping self-efficacy, and social engagement, compared to people without these disorders (Palmer *et al.*, 2014). Consequently, enhancing these traits is likely to improve well-being in a sizable proportion of people with psychiatric disorders.

The future of positive psychiatry is bright and we should look forward to its continued development and integration into the discipline of general psychiatry more broadly. This will be especially critical for geriatric psychiatry. *International Psychogeriatrics* will provide an excellent forum for publishing critically reviewed articles on positive psychiatry of aging to complement those on mental illnesses in older people.

Acknowledgments

This work was supported, in part, by the National Institute of Mental Health [NIMH R01MH094151-01 (PI: Dilip V. Jeste, MD)], and

by the Stein Institute for Research on Aging (Director: Dilip V. Jeste, MD) at the University of California San Diego.

DILIP V. JESTE

Sam and Rose Stein Institute for Research on Aging,
University of California San Diego, La Jolla, CA, USA

Email: djeste@ucsd.edu

References

- Bailly, N. et al.** (2018). Spirituality, social support, and flexibility among older adults: a five-year longitudinal study. *International Psychogeriatrics*, 30. doi: 10.1017/S1041610218000029.
- Cohen, A. N. et al.** (2017). How occupationally high-achieving individuals with a diagnosis of schizophrenia manage their symptoms. *Psychiatric Services*, 68, 324–329. doi: 10.1176/appi.ps.201600031.
- Eack, S. M. et al.** (2010). Neuroprotective effects of cognitive enhancement therapy against gray matter loss in early schizophrenia: results from a 2-year randomized controlled trial. *Archives of General Psychiatry*, 67, 674. doi: 10.1001/archgenpsychiatry.2010.63.
- Edmonds, E. C., Martin, A. S., Palmer, B. W., Eyler, L. T., Rana, B. K. and Jeste, D. V.** (2018). Positive mental health in schizophrenia and healthy comparison groups: relationships with overall health and biomarkers. *Aging & Mental Health*, 22, 354–362.
- Eglit, G. M. L., Palmer, B. W. and Jeste, D. V.** (2018). Overview of measurement-based positive psychiatry. *Nordic Journal of Psychiatry* (in press).
- Forlenza, O. V. and Vallada, H.** (2018). Spirituality, health and well-being in the elderly. *International Psychogeriatrics*, 30, 1743–1744.
- Gage, F. H.** (2002). Neurogenesis in the adult brain. *The Journal of Neuroscience*, 22, 612–613.
- Ihle, A., Oris, M., Baeriswy, M. and Kliegel, M.** (2018). The relation of close friends to cognitive performance in old age: the mediating role of leisure activities. *International Psychogeriatrics*, 30. doi: 10.1017/S1041610218000789.
- Jeste, D. V. and Palmer, B. W.** (2013). Editorial: a call for a new positive psychiatry of ageing. *British Journal of Psychiatry*, 202, 81–83.
- Jeste, D. V. and Palmer, B. W.** (eds). (2015). *Positive Psychiatry: A Clinical Handbook*. Washington, DC: American Psychiatric Publishing.
- Jeste, D. V., Palmer, B. W., Rettew, D. C. and Boardman, S.** (2015). Positive psychiatry: its time has come. *Journal of Clinical Psychiatry*, 76, 675–683.
- Mather, M. and Carstensen, L. L.** (2005). Aging and motivated cognition: the positivity effect in attention and memory. *Trends in Cognitive Sciences*, 9, 496–502. doi: 10.1016/j.tics.2005.08.005.
- Mausbach, B. T. et al.** (2007). The attenuating effect of personal mastery on the relations between stress and Alzheimer caregiver health: a five-year longitudinal analysis. *Aging & Mental Health*, 11, 637–644.

- Mohr, S. et al.** (2011). Spirituality and religiousness as predictive factors of outcome in schizophrenia and schizo-affective disorders. *Psychiatry Research*, 186, 177–182.
- Montross-Thomas, L. P., Joseph, J., Edmonds, E. C., Palinkas, L. A. and Jeste, D. V.** (2018). Reflections on wisdom at the end of life: qualitative study of hospice patients aged 58–97 years. *International Psychogeriatrics*, 3. doi: 10.1017/S1041610217003039.
- Moore, D. J. et al.** (2018). Positive psychological factors are linked to successful cognitive aging among older persons living with HIV/AIDS. *AIDS and Behavior*, 22, 1551–1561.
- Olusegun, B.** (2018). How valuable are ecopsychosocial interventions in prevention and treatment of dementia?. *International Psychogeriatrics*, 30, 1741–1742.
- Pachana, N. A. and Mitchell, L. K.** (2018). Examining the unique wisdom of older adults. *International Psychogeriatrics*, 30, 1739–1740.
- Palmer, B. W., Martin, A. S., Depp, C., Glorioso, D. K. and Jeste, D. V.** (2014). Wellness within illness: happiness in schizophrenia. *Schizophrenia Research*, 159, 151–156.
- Scelzo, A. et al.** (2018). Mixed-methods quantitative-qualitative study of 29 nonagenarians and centenarians in rural southern Italy: focus on positive psychological traits. *International Psychogeriatrics*, 30, 31–38.
- Schutte, N. S., Palanisamy, S. K. and McFarlane, J. R.** (2016). The relationship between positive psychological characteristics and longer telomeres. *Psychology and Health*, 31, 1466–1480.
- Seligman, M. E. and Csikszentmihalyi, M.** (2000). *Positive Psychology: An Introduction*. Washington, DC: American Psychological Association.
- Sharma, V. et al.** (2017). Religion, spirituality, and mental health of U.S. military veterans: results from the national health and resilience in veterans study. *Journal of Affective Disorders*, 217, 197–204.
- Summers, R. and Jeste, D. V.** (eds). (2018). *Positive Psychiatry: A Casebook*. Washington, DC: American Psychiatric Publishing.
- Wiley, J. F., Bei, B., Bower, J. E. and Stanton, A. L.** (2017). Relationship of psychosocial resources with allostatic load: a systematic review. *Psychosomatic Medicine*, 79, 283–292. doi: 10.1097/PSY.0000000000000395.

COMMENTARY

Examining the unique wisdom of older adults

When we recognize that we don't have all the time in the world, we see our priorities most clearly.

—Laura Carstensen

Montross-Thomas and colleagues begin their article “Reflections on wisdom at the end of life: qualitative study of hospice patients aged 58–97 years” with a quote from Kierkegaard on the paradox of being able to only reflect on life while continuing to live each day (Kierkegaard, 1843). The quote supplied above from Carstensen (2011) builds on this idea, suggesting that knowing that one is in the final portion of one's life can sharpen priorities and goals. Carstensen's work on future time perspective (Lang and Carstensen, 2002), in the context of her theory of socioemotional selectivity (Carstensen *et al.*, 2003), dovetails well with Montross-Thomas *et al.*'s findings on the reflections of 21 hospice patients at the end of their life. Although the context of their study was how such patients might find their perceptions of wisdom shaped by the fact of their terminal illness, the patients in this study reported that part of their challenge was to balance accepting their current circumstances, including a foreshortened future, while still striving for growth and meaning.

The participants in the current study responded to a small set of open-ended questions, which were analyzed qualitatively using a grounded theory approach. The two questions of particular relevance with respect to wisdom and the end of life were “What experiences have influenced your level of wisdom?” and “How has your illness affected your level of wisdom?” The majority of patients in the study were Caucasian (81%), male (57%), Christian (48%), and diagnosed with cancer (48%). In terms of age, the range was 58 to 97 years, with a mean age of 78 years. It is important to keep these demographics in mind when interpreting these findings, as both views of wisdom and other theories such as Carstensen's socioemotional selectivity theory have been shown to differ across cultures (e.g. Fung *et al.*, 2008). Indeed, Montross-Thomas *et al.* comment on the variation in conceptualizations of wisdom being dependent on factors including, for example, age, cohort, culture, and philosophical lens. And through their work, they aimed to establish whether

a factor such as experiencing a terminal illness might similarly contribute to the conceptualization of wisdom.

The themes that emerge from these patients' reflections on wisdom align well with available conceptualizations of wisdom, including the work of Bangen *et al.*, which the authors describe in their introduction. This is unsurprising given that in the main, the Bangen *et al.* meta-review focused primarily on research utilizing participants culturally more like those captured within the Montross-Thomas *et al.* research. Takahashi and Bordia (2000) are one of the few who have considered wisdom and culture more specifically, comparing the interpretation of wisdom by those from a Western culture with those from an Eastern culture. Results indicated more of a focus on knowledge and experience in the Western culture, while in the Eastern culture, the focus was more on aging. The examination of cultural influences remains an underdeveloped area within the wisdom literature; studies such as that of Takahashi and Bordia (2000) highlight the impact that various factors (such as culture) can have on how wisdom is conceptualized.

In addition to capturing the more common themes identified in the wisdom literature to date, Montross-Thomas *et al.* also capture one of the conundrums of the construct of wisdom, whereby individuals have their own understanding of what wisdom is, and in doing so, interpret their level of wisdom within that conceptualization. In comparison to the Bangen *et al.* meta-review, Montross-Thomas *et al.* found that terminally ill patients place similarly high importance on the wisdom themes of prosocial attitudes, behaviors, and decision making. However, the patients also ranked emotional regulation (ranked fifth in the Bangen *et al.* paper) and openness to new experience (ranked seventh in the Bangen *et al.* paper) as higher in importance. Reflection and self-understanding and value relativism and tolerance were ranked lowest, while in the Bangen *et al.* meta-review, the former component was ranked third overall and the latter ranked sixth. Therefore, the link between Carstensen's socioemotional selectivity theory is appropriate, with more consideration given to meaningful social ties, to the

sharing of self with others, and a greater appreciation of what is left in life by those experiencing a terminal illness. This is then reflected in what participants see as the most pertinent elements of wisdom, namely those more focused on the meaningful social elements of their lives.

Of course, it is normal research practice to aim to compartmentalize and make qualitative data more meaningful by grouping it into themes. And so, a refreshing element of Montross-Thomas *et al.*'s work is that they also look to consider the unique insights offered by this group of patients. This work therefore highlights not only the common themes that seem to resonate fairly consistently within Western-based cultures in particular (Mitchell *et al.*, 2017), but also highlights that wisdom continues to contain an element that is individually driven, likely aligned with the values, beliefs, and philosophical underpinnings of the individual themselves, as well as their own wisdom development pathway (Glück and Bluck, 2013).

This individually formulated perspective on the end of life and one's attitude toward death may be related to what Monk (2014) calls adaptive resignation, or the gradual recognition that the circumstances of one's life are changing such that time is running out, similar to Carstensen's ideas about changing future time perspectives (Lang and Carstensen, 2002). This in turn is related to positive actions at the end of life, such as wanting to pass on knowledge and wisdom to future generations, which may also have therapeutic value in the form of life lessons written and passed on, utilized as part of interventions for those in palliative care (Kasl-Godley and Christie, 2014).

Finally, Montross-Thomas *et al.* offer a conceptual model of wisdom aligned with terminally ill patients that focuses on the balance between two elements: active acceptance and galvanized growth. These two elements are an intrinsic part of the challenge of the end of life experience, as reflected on in the article: "... wisdom involved a delicate balance between learning to simply be, while continually striving to change" (p. 6). In this way, the authors capture the lens through which terminally ill patients see wisdom and the role wisdom plays within their lives, knowing that there is an end in sight.

Although in one sense, palliative care has been practiced for centuries, attitudes and approaches to death and dying have shifted, and continue to shift, over time. Thus, current best practice in palliative care remains an area which is deserving of continued research. There are new models emerging which take into account both the need for specialist palliative care practitioners, as well

as knowledge of the care for such patients into generalist practice, as well as training the next generation of healthcare professionals to recognize key components of best practice palliative care (Quill and Abernethy, 2013). Studies such as that of Montross-Thomas and colleagues brings another perspective on such patients, beyond easing suffering at the end of life, encompassing what that life, and its approaching end, contribute to reflections on a range of human values, including wisdom.

N. A. PACHANA AND L. K. MITCHELL

University of Queensland, Brisbane, Australia

Email: npachana@psy.uq.edu.au

References

- Carstensen, L. L.** (2011). *A Long Bright Future*. New York, NY: PublicAffairs.
- Carstensen, L. L., Fung, H. H. and Charles, S. T.** (2003). Socioemotional selectivity theory and the regulation of emotion in the second half of life. *Motivation and Emotion*, 27, 103–123.
- Fung, H. H., Stoeber, F. S., Yeung, D. Y.-I. and Lang, F. R.** (May 2008). Cultural specificity of socioemotional selectivity: age differences in social network composition among Germans and Hong Kong Chinese. *The Journals of Gerontology: Series B*, 63, P156–P164.
- Glück, J. and Bluck, S.** (2013). MORE wisdom: a developmental theory of personal wisdom. In M. Ferrari and N. Weststrate (eds.), *The Scientific Study of Personal Wisdom* (pp. 75–98). New York, NY: Springer.
- Kasl-Godley, J. E. and Christie, K. M.** (2014). Advanced illness and the end of life. In N. A. Pachana and K. Laidlaw (eds.), *Oxford Handbook of Clinical Geropsychology* (pp. 144–160). Oxford, UK: Oxford University Press.
- Kierkegaard, S.** (1843). Journalen JJ. In *Søren Kierkegaard's Skrifter* (vol. 18, p. 306). Copenhagen: Søren Kierkegaard Research Center.
- Lang, F. R. and Carstensen, L. L.** (2002). Time counts: future time perspective, goals, and social relationships. *Psychology and Aging*, 17, 125.
- Mitchell, L. K., Knight, B. G. and Pachana, N. A.** (2017). Wisdom across the ages and its modern day relevance (editorial). *International Psychogeriatrics*, 29, 1231–1234.
- Montross-Thomas, L. P., Joseph, J., Edmonds, E. C., Palinkas, L. A. and Jeste, D. V.** (2018). Reflections on wisdom at the end of life: qualitative study of hospice patients aged 58–97 years. *International Psychogeriatrics*, doi: 10.1017/S1041610217003039.
- Munk, K. P.** (2014). Transitions in later life. In *Oxford Handbook of Clinical Geropsychology* (pp. 144–160). Oxford, UK: Oxford University Press.
- Quill, T. E. and Abernethy, A. P.** (2013). Generalist plus specialist palliative care – creating a more sustainable model. *New England Journal of Medicine*, 368, 1173–1175.
- Takahashi, M. and Bordia, P.** (2000). The concept of wisdom: A cross-cultural comparison. *International Journal of Psychology*, 35, 1–9.

COMMENTARY

How valuable are ecopsychosocial interventions in prevention and treatment of dementia?

Dementia is a disorder that arouses major public health interest and concern. It has been projected that there will be a global increase in the number of people affected from about 46.8million in 2018 to 131million by 2050; global cost of care for 2015 was put at US\$818 billion (Prince *et al.*, 2015). Consequently, such development will lead to tremendous social and financial cost on family and society. Currently, there is no cure for dementia and that has led to increased research activities on prevention strategies, which often has to start with a number of midlife activities. These include regular exercise, diet, treatment of cardiovascular risk factors, and social and educational stimulation through life.

Cognitive decline is a major aspect of dementia; indeed, it usually predates its onset. In both prevention and treatment of dementia reducing the rate of cognitive decline is a major investment in delaying onset or limiting severity. There are two major ways of treating dementia and these are broadly referred to as pharmaceutical and non-pharmaceutical. Pharmacological treatment broadly refers to use of cholinesterase inhibitors and memantine; of course, there is the hope for introduction of anti- amyloid drugs in the near future. Non-pharmacological treatment methods refer to psychoeducation, cognitive training, exercise, and leisure activities among others, some of which have cultural coloration. The term non-pharmacological intervention has been criticized as not being specific enough by Zeisel *et al.* (2016) who went on to suggest ecopsychosocial intervention.

In this paper, *The relation of close friends to cognitive performance in old age: the mediating role of leisure activities* (Ihle *et al.*, 2018b) report that having close friends enhances cognitive reserve through leisure activities and may help preserve cognitive function in older adults. Previously, the group had reported on cognitive reserve, social capital, and cognitive performance in old age (Ihle *et al.*, 2018a). Markers of cognitive reserve here are education, cognitive level of job, and leisure activities, while markers of social capital are numbers of significant family members and number of close friends. In another report, *The association*

of educational attainment, cognitive level of job, and leisure activities during the course of adulthood with cognitive performance in old age: the role of openness to experience. Ihle *et al.* (2016) reported that higher education level, higher cognitive job experience, higher number of leisure activities, and higher score on openness were significantly associated with better cognitive performance. Openness in this context is a measure of personality dimension from the Big Five Inventory (Rammstedt and John, 2007). Engaging in leisure activities was greater in individuals with high openness to experience and individuals with higher openness experience also showed better cognitive performance (Ihle *et al.*, 2016). It would appear that certain other variables could influence selection of leisure activities and even influence education and job selection. In the current paper, individuals show greater engagement in leisure activities if they have large number of close friends. This finding remains robust after adjusting for potentially confounding variables like education, marital status, retirement status, gender, and subjective health status. This is a cross-sectional study which has its limitations; however, the result of one longitudinal study differs somehow. Hakansson *et al.* (2009) in a prospective population study with mean follow-up time of 21 years observed that people in marriage or living with a partner in midlife (mean age 50 years) were less likely to have cognitive impairment at ages between 65 and 79 years when compared with those divorced, widowed, separated, or never married. It is intriguing that in their study, marriage protects against cognitive decline. In the present study, it does not appear that robust. It could be argued that the closest friend an individual has in a marriage that is free of rancor is his or her spouse. The authors admit some limitations in the present study which include possible selection bias, in that, participants in the study were expected to complete a test of vocabulary ability as well as trail making tests. Participants who fail to complete these tests were not included in analysis; thus, it is possible that those who already have some impairment of cognitive abilities might have been excluded; however, the study aimed to recruit cognitively normal individuals only.

Despite the limitations, the public health implications of the study are enormous, for both treatment (non-pharmacological) and prevention strategies in dementia. It could be presumed that having close friends and engaging in leisure activities should at least be preventive of cognitive decline in older adults who are currently not married. In a review by Larson (2010), there was emphasis on compression of cognitive morbidity as a strategy for delaying the onset of Alzheimer's disease by extension limiting Disability Life Years in persons with dementia, as it is conceivable that some may die of causes other than dementia as age advances before dementia sets in. In an earlier review (Fratiglioni *et al.*, 2004), it was pointed out that higher levels of cognitive functioning could be due to leisure activities that are stimulating and that leisure activities correlated negatively with cognitive decline. Leisure activities may include attending cultural events, music, singing, reading books, or magazines and solving cross word puzzles. Lautenschlager *et al.* (2008) in a randomized control trial, reported that physical exercise protected persons with mild cognitive impairment from further cognitive decline when compared with usual treatment group; the difference remained for 18 months after the trial was over. More importantly, the effect size due to exercise was significantly larger than that of cholinesterase inhibitors.

In conclusion, it would appear that the role of close friends and leisure activities in relation to cognitive performance can only be part of a whole and not the whole in the process of enhancing cognitive performance or delaying cognitive decline. Zeisel *et al.* (2016) commented that because of the poor definition of non-pharmacological treatment method (which they likened to a state of non-hate rather than love), not much attention has been paid to this treatment or prevention arm of dementia when compared with drug development, as a matter of fact non-pharmacological treatment methods receive relatively poorer funding. Efforts in this area as shown by the findings in this paper will most likely bring the desired change.

Conflict of interest

None.

OLUSEGUN BAIYEWU

Department of Psychiatry, College of Medicine,
University of Ibadan, Ibadan, Nigeria
Email: olusegun_baiyewu@yahoo.com

References

- Fratiglioni, *et al.*** (2004). An active and socially integrated lifestyle in late life might protect against dementia. *Lancet Neurology*, 3, 343–353.
- Hakansson, K. *et al.*** (2009). Association between mid-life marital status and cognitive functions in later life: population based cohort study. *BMJ*, 339, b2462. doi:10.1136/bmj.b2462.
- Ihle, A. *et al.*** (2016). The association of educational attainment, cognitive level of job, and leisure activities during the course of adulthood with cognitive performance in old age: the role of openness to experience. *International Psychogeriatrics*, 28, 733–740.
- Ihle, A. *et al.*** (2018a). Cognitive reserve and social capital accrued in early and midlife moderate the relation of psychological stress to cognitive performance in old age. *Dementia and Geriatric Cognitive Disorders*, 45, 190–197.
- Ihle, A. *et al.*** (2018b). The relation of close friends to cognitive performance in old age: the modulating role of leisure activities. *International Psychogeriatrics*, doi: 10.1017/S1041610218000789.
- Larson, E. B.** (2010). Prospects for delaying the rising tide of worldwide late-life dementias. *International Psychogeriatrics*, 22, 1196–1202.
- Lautenschlager, N. T. *et al.*** (2008). Effect of physical activities on cognitive in older adults at risk for Alzheimer's disease: a randomized trial. *JAMA*, 300, 1027–1037.
- Prince, M. *et al.*** (2015). *World Alzheimer report. The global impact of dementia: an analysis of prevalence, incidence, cost and trends.* Alzheimer's Disease International: London.
- Rammstedt, B. and John, O. P.** (2007). Measuring personality in one minute or less: a 10 item version of big five inventory in english and German. *Journal of Research and Personality*, 41, 203–212.
- Zeisel, J. *et al.*** (2016). Ecopsychosocial interventions in cognitive decline and dementia: a new terminology and a new paradigm. *American Journal of Alzheimer's Disease & Other Dementias*, 31, 502–507.

COMMENTARY

Spirituality, health and well-being in the elderly

Bailly *et al.* (2018) examined the trajectory of spirituality in a cohort of 567 non-institutionalized older adults living in Tours, France, during a period of five years. The measurements for spirituality (Daily Spiritual Experience Scale, DES), social support (Satisfaction with Social Support subscale of the Duke Social Support Index), and accommodative tendencies (Flexible Goal Adjustment) were longitudinally collected at three time points (2007, 2009, and 2012). The results of the study confirmed some expected observations, such as higher levels of spirituality among religious older adults when compared with the ones without religion, and older women reporting higher levels of spirituality than older men. But the most interesting finding was the observation that the measured levels of spirituality among older adults remained stable during this five-year period. Based on a growing number of studies and theories of aging suggesting that the levels of spirituality increases during a person's lifetime, one would expect an increase in the levels of spirituality along the study follow-up. The authors, however, interpreted the stable level of spirituality informed by the participants as having already reached a relatively high mean rating score of spirituality at baseline. From the beginning of the trail, many participants expressed self-contentment and reported having found meaning in their lives. Moreover, the responders had, in general, more years of education than expected for people in their age group, lived at home independently with a good self-health evaluation, had relatively fewer diseases, and a good perception of financial satisfaction; these characteristics perhaps make this group not representative of the French general population in the same age bracket.

One of the factors related to changes in attitudes leading to a spiritual transcendence is the transformation of the environment and/or health status. Most of the publications on spirituality investigate the relationship with health and the levels of spirituality in the context of a patient's disease. As the authors rightly commented, more than age *per se*, it may be the way age-related changes in health functioning, social losses, and approaching death are dealt with that could be one of the relevant factors for the development

of spirituality among older adults. For example, the respondents of this study continue having a relatively stable daily routine and *modus vivendi* and presumably even in difficult times.

Spirituality and health (mental and physical) is a fascinating area that started to develop as a scientific field only at the beginning of the XXI century (Koenig, 2008). Bailly's original cohort is an example of it. Their cohort started in 2001, with evaluations every two years, but only included the measuring of spirituality, in 2007, missing these measurements in the assessment waves of 2001, 2003, and 2005. The growing interest in this area is illustrated by the increasing number of publications reporting a positive correlation between spirituality and better outcomes in prevention, healing, and coping with diseases. At the same time, we still know very little about the mechanisms that underlie these interactions.

Another important point raised by Bailly and her coauthors concerns the definition of spirituality. The traditional–historical meaning defines a “spiritual person” as a deeply religious individual, but in the last 20–30 years the definition of spirituality became quite a broad and diffuse term that differs according to the individual user, and may signify, for instance, the superficially religious person, the religious seeker, the seeker of well-being and happiness, or the completely secular person (Koenig, 2008). As a consequence of this “polymorphic” characterization of spirituality, there is little agreement today within the academic community upon a common definition for spirituality. A number of scales, indexes, and questionnaires have been developed to measure spirituality in research. However, those measurements from research data reflect different concepts and therefore lead to different valuations with limited overlap between them, making the comparison of the results of two independent studies many times very difficult or even impossible. As an example, the DES used by Bailly *et al.* in this paper measures a very broad concept of spirituality, which includes measures of peacefulness, harmony, and other positive experiences. However, with this definition, an individual with depression or anxiety who rarely has such feelings of harmony and peacefulness would have difficulties to identify with it. Again,

when the authors compare their results with another rare longitudinal study (Wink and Dillon, 2003), they found out that both studies used a distinct method for measuring the levels of spirituality and, therefore, the results are not readily comparable. Wink and Dillon (2003) used answers to open-ended questions for spirituality, which were then coded between 1 and 5, and later those ratings were validated against the Religion Index for Psychiatric Research.

The current problem that arises from not having a consensus definition for spirituality could be compared to the difficulties that researchers had to classify psychiatric disorders before the development of operational criteria in psychiatry, in the 1960's and 1970's, when it was difficult to compare the results of clinical studies in a variety of psychiatric disorders. Such a methodological development is also required in the field of spirituality. However, there is an advantage from the clinical perspective to use a broad concept of spirituality, since it also expands eligibility, i.e. every person is more or less spiritual – including atheists. Patients can define whatever spirituality means to them and the health worker will address it according to each patient's definition. Nonetheless, the use of this universal characterization would make research on the relationship between spirituality and health very difficult, since all participants will be spiritual (but to different degrees) (Peteet *et al.*, 2018). An alternative definition of the term spirituality to be used in research, according to Koenig, would be to apply it only to a subgroup of deeply religious people: the ones who have dedicated their lives to the service of their religion and to their fellow humans, and whose lives exemplify the teachings of their faith and traditions (Koenig, 2008). This narrow definition would lead to the exclusion of those groups of people who still consider themselves spiritual but do not lead such spiritually dedicated lives. To include the term religion, on the other hand, has its advantages: is a more distinct construct with multiple dimensions, which that can be measured and distinguished from one another; here is very little disagreement between investigators on the definition of this term.

In conclusion, the need for more studies is clear, especially to understand the effects and

mechanisms of action/relationship of spirituality and health/well-being. Despite there being few methodologically sound studies – particularly, longitudinal ones like the study presented here by Bailly *et al.* – it would be very welcome (and challenging) for investigators to reach a common denomination for spirituality, i.e. to have a consensual agreement in order to establish a set of core or minimal standardized measurements of spirituality to be used in future research, enabling the comparison of results across different groups, settings, and cultures. In this way, it would be possible to shorten the time and lessen the resources to identify the possible mechanisms of action of this proposal.

ORESTES V. FORLENZA¹ AND HOMERO VALLADA²

¹Laboratório de Neurociências (LIM-27), Instituto de Psiquiatria, Hospital das Clínicas HCFMUSP, Faculdade de Medicina, Universidade de Sao Paulo, Sao Paulo, SP, Brazil

²Laboratório de Psicopatologia e Terapêutica Psiquiátrica (LIM-23/ProSER), Instituto de Psiquiatria, Hospital das Clínicas HCFMUSP, Faculdade de Medicina, Universidade de Sao Paulo, Sao Paulo, SP, Brazil

Email: forlenza@usp.br

References

- Bailly, N. et al.** (2018). Spirituality, social support, and flexibility among older adults: a five-year longitudinal study. *International Psychogeriatrics*, doi: 10.1017/S1041610218000029.
- Koenig, H. G.** (2008). Concerns about measuring “spirituality” in research. *Journal of Nervous and Mental Disease*, 196, 349–355. doi: 10.1097/NMD.0b013e31816ff796.
- Peteet, J. R., Zaben, F. A. and Koenig, H. G.** (2018). Integrating spirituality into the care of older adults. *International Psychogeriatrics*, doi: 10.1017/S1041610218000716.
- Wink, P. and Dillon, M.** (2003). Religiousness, spirituality, and psychosocial functioning in late adulthood: findings from a longitudinal study. *Psychology and Aging*, 18, 916–924.

Reflections on wisdom at the end of life: qualitative study of hospice patients aged 58–97 years

Lori P. Montross-Thomas,^{1,2,3} Jamie Joseph,² Emily C. Edmonds,²
Lawrence A. Palinkas⁴ and Dilip V. Jeste^{2,3,5}

¹Department of Family Medicine & Public Health, University of California San Diego, San Diego, California, USA

²Department of Psychiatry, University of California San Diego, San Diego, California, USA

³Sam and Rose Stein Institute for Research on Aging, University of California San Diego, San Diego, California, USA

⁴Department of Children, Youth, and Families, University of Southern California School of Social Work, Los Angeles, CA, USA

⁵Department of Neurosciences, University of California San Diego, San Diego, California, USA

ABSTRACT

Objective: Wisdom is a complex trait, and previous research has identified several components of wisdom. This study explored the possible impact of a diagnosis of a terminal illness on the conceptualization and evolution of wisdom while facing the end of life.

Design and Participants: Semi-structured qualitative interviews were conducted with 21 hospice patients aged 58–97 years who were in the last six months of their life.

Methods: Hospice patients were asked to describe the core characteristics of wisdom, as well as how their terminal illness might have impacted their understanding of this concept. The interviews were audiotaped, transcribed, and coded by the research team using a grounded theory analytic approach based on coding consensus, co-occurrence, and comparison.

Results: Broad concepts of wisdom described by the hospice patients align with the extant literature, thereby supporting those general conceptualizations. In addition, hospice patients described how their life perspectives shifted after being diagnosed with a terminal illness. Post-illness wisdom can be characterized as a dynamic balance of actively accepting the situation while simultaneously striving for galvanized growth. This delicate tension motivated the patients to live each day fully, yet consciously plan for their final legacy.

Conclusion: The end of life offers a unique perspective on wisdom by highlighting the modulation between actively accepting the current situation while continuing the desire to grow and change at this critical time. This paradox, when embraced, may lead to even greater wisdom while facing one's own mortality.

Key words: palliative care, cancer, spirituality, resilience

“Life can only be understood backwards; but it must be lived forwards.”

— Søren Kierkegaard, Philosopher (1843)

Introduction

While wisdom is an ancient religious and philosophical concept (Takahashi, 2000; Jeste and Vahia, 2008), one of the first modern scientists to consider wisdom as a psychological construct was Erikson (1950). He described foundational stages

of psychosocial development, with the final (eighth) stage being characterized by a conflict between ego integrity and despair; the optimal resolution of this stage was acquisition of wisdom. Empirical research on wisdom began in the 1970s and has been growing in recent years (Clayton, 1975; Baltes and Smith, 2008). Modern conceptualizations describe wisdom as an aggregate and inter-related mix of cognitive, affective, and reflective attributes (Sternberg, 1990; Ardelt, 2004; Thomas *et al.*, 2017).

To develop a consensus definition of wisdom based on the published literature, Bangen *et al.* (2013) conducted a meta-review utilizing cohorts ranging from adolescence (Damon, 2000; Pasupathi *et al.*, 2001; Lerner, 2008) to older adulthood (Smith and Baltes, 1990; Happé *et al.*, 1998; Glück

Correspondence should be addressed to: Dilip V. Jeste, M.D., University of California San Diego, 9500 Gilman Drive, Mail Code #0664, La Jolla, California 92093-0664, USA. Phone: +1-858-534-4020. Email: djeste@ucsd.edu. Received 21 Sep 2017; revision requested 21 Nov 2017; revised version received 27 Nov 2017; accepted 9 Dec 2017. First published online 24 January 2018.

et al., 2005). The review integrated conceptualizations from 24 empirical definitions across 31 articles, and identified 9 distinct components of wisdom, listed in decreasing order of frequency of citations in published definitions: (1) social decision making and general knowledge of life, (2) prosocial attitudes and behaviors, (3) reflection and self-understanding, (4) acknowledgement of uncertainty, (5) emotional regulation, (6) value relativism and tolerance, (7) openness to new experience, (8) spirituality and religiosity, and (9) sense of humor.

Although this meta-review provided cohesion for wisdom conceptualizations, the concept of wisdom may vary somewhat, depending on age-based, cultural, contextual, historical, philosophical, or religious lenses used to investigate the construct (Takahashi, 2000; Jeste and Vahia, 2008; Kross and Grossmann, 2012). In terms of lifespan, hospice care has been redefining our understanding of the affective, cognitive, physical, social, and spiritual underpinnings of well-being as death draws near (Pinder and Hayslip, 1981; Murray *et al.*, 2007; Vachon *et al.*, 2009). Most notably, it has been suggested that the end of life can provide a unique time for possible self-transcendence and self-reflection (Vachon *et al.*, 2009), constructs often associated with the presence of intelligence, spirituality, and wisdom (Jeste *et al.*, 2010). Facing mortality is thought to evoke a greater awareness about larger life perspectives (Bellizzi, 2004), and a dynamic relationship can exist between positive and negative life experiences at this stage (Carstensen *et al.*, 1999; Diamond and Aspinwall, 2003; Meier, *et al.*, 2016). Therefore, assessing individuals at the end of their lives may be particularly salient for characterizing and understanding their perspectives on wisdom.

To explore this possibility, we qualitatively interviewed patients who were diagnosed with a terminal illness and receiving hospice care during the last six months of their life. We examined their reflections regarding the characteristics and influences of wisdom. The study aimed to understand whether a hospice cohort could offer a distinct perspective on wisdom, given their potential to uniquely reflect back on life, coupled with their current understanding of how to live life when faced with a terminal illness.

Methods

Procedures for data collection and analysis were reviewed and approved by the Human Research Protections Program at the University of California San Diego, USA. Informed consent was obtained

from each hospice patient after the study objectives had been fully explained.

Patients were recruited by members of their hospice team with informational study brochures. The study inclusion criteria were purposely broad, given the novel and exploratory nature of this research: all hospice patients who were English speaking and able to complete a one-hour interview were eligible to participate. Study recruitment remained open until the data evidenced theoretical saturation (Morse, 1995).

After study enrollment, each patient completed an individual, 60-minute, semi-structured interview with a mental health professional working in hospice care. All the interviews were audiotaped and conducted in the patients' homes or residences. The interviews were conducted using a semi-structured interview guide, which included questions regarding the descriptions and characteristics of wisdom as well as life influences on wisdom. Primary questions in the interviews included: "How do you define wisdom?," "What are the characteristics of wise people, in your opinion?," "What experiences have influenced your level of wisdom?," and "How has your illness affected your level of wisdom?" Notably, all the interviews were open-ended to allow patients to introduce or expand upon topics of importance to them and to allow for additional follow-up questions, as needed, to learn more about each patient's unique perspective.

Upon completion of the interviews, each audiotape was transcribed, providing a total of 236 pages for analysis (the average patient interview was 11.2 pages in length). These transcripts reflected the content of the interviews verbatim, thereby allowing reviewers to code the content in full. Based within the methodology of "Coding Consensus, Co-occurrence, and Comparison" as outlined by Willms *et al.* (1990), the interview transcripts were analyzed using a grounded theory analytic approach (Glaser and Strauss, 1967).

First, approximately one-quarter (24%, $n = 5$) of the interviews were randomly selected for coding by the first and second authors. This coding was completed at a general level by identifying common words or phrases seen in the interviews, in order to condense the data into analyzable units. Paragraphs within each interview were assigned open and axial codes based on *a priori* (i.e. questions in the interview guide) or emergent themes (Strauss and Corbin, 1998). In many instances, the same paragraph was assigned multiple codes. These initial codes were provided to the full research team. Disagreements in assignments or descriptions of codes were resolved through a discussion among all the study authors and a final coding matrix was created.

Next, the first and second authors independently coded all 21 interviews in their entirety using the agreed-upon coding matrix. These two authors then reviewed all the codes proposed and discussed whether they agreed or disagreed with each of the codes as chosen by the other. Comparison of codes assigned to one of the transcripts revealed an inter-rater reliability rate of 96% – a level indicating high concordance between coders (Boyatzis, 1998).

Finally, all of the codes were entered into Dedoose, a computer program designed for qualitative and mixed-methods analyses (Dedoose, 2015). Using the method of constant comparison (Glaser and Strauss, 1967), codes were then grouped into broader themes, and provided the basis for a group discussion among all the authors regarding the thematic salience and ensuing conceptual map of themes.

Results

Participants

Twenty-one patients diagnosed with a terminal illness, who had a prognosis of six months or less to live, were enrolled in the study between 2012 and 2015. The patients were receiving care at either San Diego Hospice or Mission Hospice, both located in San Diego, California, USA. Patient ages ranged from 58 to 97 years, with a mean age of 78 years ($SD = 11$). A majority of the patients were Caucasian (81%, $n = 17$) and approximately one-half were male (57%, $n = 12$), Christian (48%, $n = 10$) and diagnosed with cancer (48%, $n = 10$). The remaining diagnoses included chronic obstructive pulmonary disease, congestive heart failure, amyotrophic lateral sclerosis, pulmonary fibrosis, and end-stage liver disease. Most study patients were either widowed (38%, $n = 8$) or currently married (33%, $n = 7$).

Descriptions and characteristics of wisdom

In response to open-ended questions regarding general characteristics of wisdom, patients spontaneously discussed each of the nine components of wisdom as previously identified in the meta-review by Bangen *et al.* (2013), thereby supporting the review's overall findings. However, the order of thematic salience of individual components for this hospice sample (Table 1) differed from that based on salience in the published literature (given above in the Introduction). For hospice patients, the order was: (1) prosocial attitudes and behaviors, (2) social decision making and general knowledge of life, (3) emotional regulation, (4) openness to new experience, (5) acknowledgment of uncertainty, (6)

spirituality and religiosity, (7) reflection and self-understanding, (8) sense of humor, and (9) value relativism and tolerance. See Table 1 for exemplar patient quotes related to each general theme.

Influences of Illness on Wisdom

IMPACT OF ILLNESS AND RECEIVING HOSPICE CARE

In addition to the general conceptualizations of wisdom presented above, the hospice patients in this sample were able to provide additional unique insights into how wisdom may build or transform after being diagnosed with a terminal illness. These shifts were not necessarily changes the patients had intentionally cultivated. Instead they described dramatic shifts that occurred in a relatively short time frame, and ones that were directly linked to being seriously ill and/or initiating hospice care.

“I think I have learned wisdom, but I did not learn it from home. I have learned most of it from being in the situation I am in now, really. I never thought about it before. It was just, you know... I was doing my own thing and everything was fine. Then you get to a point where you cannot do it anymore, and the wisdom comes in learning how to handle the situation you are in.”

“I really did not have to think about wisdom before I was ill.”

“I guess that's the last experience that I've had to deal with. My perspective, my outlook on life - my outlook on everything - has changed. It's grown tremendously.”

ACTIVE ACCEPTANCE

Patients described their hope of finding a sense of acceptance or peace related to their illness, particularly in light of their ensuing physical changes and loss of functioning. This theme was not described as a passive “giving up,” but as an active coping process. Here patients emphasized appreciating life, taking the time to reflect, and even finding ways to live more simply than before they became ill. There was also a keen sense of fully enjoying the time they had left and in so doing, finding the beauty in everyday life.

“For all my life, being a Southerner and having been in beauty contests, I got up in the morning, put my full makeup on, and did my hair every day. A lady was never in her night gown unless she was giving birth! Now all that is very, very difficult for me and I think that has been really hard. I've accepted it, and I realized that I have to let it go. I have to ask for help and allow them to help me. I try to take all this with as much graciousness as possible - which I've learned is

Table 1. Emergent themes reported by 21 hospice patients regarding general descriptions and characteristics of wisdom

| EMERGENT THEME | EXEMPLAR QUOTES |
|--|--|
| Prosocial attitudes and behaviors | <i>"I think you would have more wisdom if you have empathy and compassion. Because with empathy and compassion, comes understanding."</i> <i>"I've never seen anybody who is self-centered who I can say is wise."</i> |
| Social decision making and general knowledge of life | <i>"I think a wise person goes and seeks counsel and looks for information before they just jump in and make a decision... They weigh the consequences and the pros and the cons."</i> <i>"Wise people, in my opinion, will think a great deal before they make any judgments."</i> |
| Emotional regulation | <i>"I think many times your emotions and personal problems can get in the way of being wise."</i> <i>"Every day is a living experience, and it is what you can make of it, whether it is going to be a happy day or not. What you allow into your life makes you happy or sad."</i> |
| Openness to new experience | <i>"Wisdom is knowledge learned by your mistakes and the action you take to correct those mistakes."</i> <i>"Keep involved with things. Keep your eyes open, your ears open, study, learn all you can about every subject, and do not stop. Maybe then you will be wise..."</i> <i>"I think that's how we gain wisdom - by experience and observation and having an open mind to everything that comes along."</i> |
| Acknowledgement of uncertainty | <i>"There is no way to know it all, that's the whole problem. You don't know it all. Nobody will, and I think maybe that's wisdom in itself - realizing that you don't know anything."</i> <i>"Wisdom means seeing life on life's terms."</i> |
| Spirituality and religiosity | <i>"I ask for wisdom... I have a relationship with God, and if I need something, I ask Him."</i> <i>"To me, wisdom is not from man. I get my knowledge and understanding from above."</i> <i>"I feel I'm wise every time I let go and surrender my little ego self; all you really have to do is plant the seed, and then get out of the way and let the universe produce for you."</i> |
| Reflection and self-understanding | <i>"Start getting to know yourself... Because everything you're ever gonna need, you've already got."</i> <i>"Wisdom is the inner voice that we're given at birth... but most people seem to seek exterior forces and people, places, and things in order to make them happier, but to me wisdom itself emanates from within us, we just have to slow down long enough to find it."</i> |
| Sense of humor | <i>"There is usually humor in a lot of things although there is sadness too. You cannot listen to that sadness. You have to get out of it, or you get so depressed, you know. Then you are not good to anyone and you are absolutely useless to yourself."</i> |
| Value relativism and tolerance | <i>"I think the characteristics of wise people are people who listen. They are good listeners... and they aren't judgmental. They are willing and open to listening to all sides before they say anything."</i> |

Note: Themes listed in order of thematic salience based on the hospice patients' interviews. Themes compared to the components of wisdom as described in the meta-review by Bangen *et al.* (2013).

more important anyway. And I realized that my friends really don't care. They don't care that I don't have makeup on or I'm in my night gown. They are just happy to see me out of bed sitting in a chair."

"Well, I do not think I am the wisest person, but now I think wisdom is about cultivating a happy attitude in your life. Not necessarily based on having money, but being happy with just looking at the sky and appreciating nature and loving the people around you. I think then you will have a very rich life."

"Unfortunately, my body does not keep up with my mind. I'm limited in the things I'd like to do and want to do, but at the same time you have to make adjustments. I mean, I used to be an avid tennis player and hiking was a part of my life. I've had to say, 'Well that's behind you.' I am very fortunate to have

a daughter and a wife who take me out places and who take me to watch the seagulls at sunset."

"Hmmm...what has changed? Well, one thing is that I have no interest in money. Money used to be pretty important... I always thinking 'What could I buy?' or 'What could I do?' I am not that way anymore. Nothing material is important since I have been on hospice."

"I know when I moved here it was the best decision I could have made. It was a painful one and then I realized it was a wise decision to stop treatment. I made the decision before it had to be made for me. You can spin your wheels and go from doctor to doctor looking for hundreds of different treatments... But there comes a day when you have to accept the fact that everything that can be done has been done."

GALVANIZED GROWTH

Patients also spoke of positive changes they encountered in response to their illness. These adaptive characteristics were stimulated and forged by the difficulty of living with a terminal illness, and this galvanizing process was linked directly to increased wisdom. For example, most patients talked about finding increased determination, gratitude, positivity, and strength. Patients further noted increases in spiritual or religious practices as they connected to these evolving aspects of themselves.

“Every day I wake up and I am alive, I am thankful. I do not take it for granted. I did before I got sick - but not now.”

“I think I’m a little stronger in my life. When you are with the disease, it makes you have a lot of patience.”

“I think I know more about people; people’s reactions, what people are really striving for, and the importance of goals and things that I did not even think about before.”

“I say to the Lord when I wake up in the morning, ‘My God, thank you for giving me my new life.’ Because when you get up in the morning, it is a new life. Someday I will not get up no more.”

“Now wisdom is being aware of my surroundings, trying to read the people that I meet, and trying to appreciate my day and look for the gifts. Look for the positive instead of the negative, I would say.”

WISDOM INVOLVES PERSISTENT AND DYNAMIC BALANCE

Ultimately, patients described wisdom as a perpetual balance between actively accepting illness on one hand, while wanting to grow and change on the other (see Figure 1 for a conceptual model). Contentment was seen when both sides of the “seesaw” experience were acknowledged, with an understanding that on some days the seesaw forgivably tipped more to one side than the other. There was no “perfect” solution in the face of illness, but rather on-going vacillations between learning to accept things as they were versus striving to change. For patients, wisdom involved honestly recognizing this struggle and humbly allowing it to exist. This process often led to clearer intentions about how the patients wanted to live during their remaining days, with a sense of gravitas for how those final days would directly reflect their life’s legacy.

“I started a new business, kept that going, and then I guess the last and final life-changing event was being diagnosed with stage IV lung cancer - inoperable and

basically terminal. I’m working on living as long as I can, but the point is that I am looking forward to taking whatever God has given me, whatever I have during the rest of my life, then I will leave it.”

“I’ve got three more treatments, then we’re gonna do the testing and all of that. We will see how it all works up. But whatever way it works, it’s just gonna work out the way it is supposed to. I’ll do what I can.”

“I had to learn how to do nothing – I went from a person that did everything to a person that does nothing. It takes more wisdom than I had. I have it now because I have learned how to cope with it, and I am more comfortable with my situation. Not 100%, but more comfortable.”

“Well, right now, I could just sit around and feel sorry for myself and not do anything at all. And what a waste that would be at the end of my life.”

“I want them to remember me with a smile, laughing and giggling and doing some of the silly things we do. You know, it is fun. Why do you want to leave on a sad note? I do not want to be remembered being sad.”

“You know I’m terminal. When I came in, they said there will be two or three weeks... it has been six weeks, and now it is getting to the point where my ability to swallow is gone; it should happen today or tomorrow. I know I’m going [to die] and this is wisdom. I’m not going to worry about whether I chose the right thing to do, or whether my life was good enough. I know I’m going... and I don’t fear.”

Discussion

This study offers a unique perspective on wisdom from patients receiving hospice care at the end of their lives, thereby contributing to previous conceptualizations of wisdom’s cognitive, reflective, and affective origins (Clayton and Birren, 1980; Ardel, 2004; Thomas *et al.*, 2017). There are several similarities between the themes discussed by this hospice sample and those previously reported in the literature. Specifically, nine of the subcomponents of wisdom spontaneously described by these hospice patients were similar to those outlined in the meta-review by Bangen *et al.* (2013); however, the order of salience was different. Thus, compared to the frequency of citations in the published papers, the dying patients ranked emotional regulation, openness to new experience, and spirituality higher, whereas reflection and self-understanding, and value relativism and tolerance were ranked lower.

Patients in this study explained the nuances of how receiving a terminal illness and entering

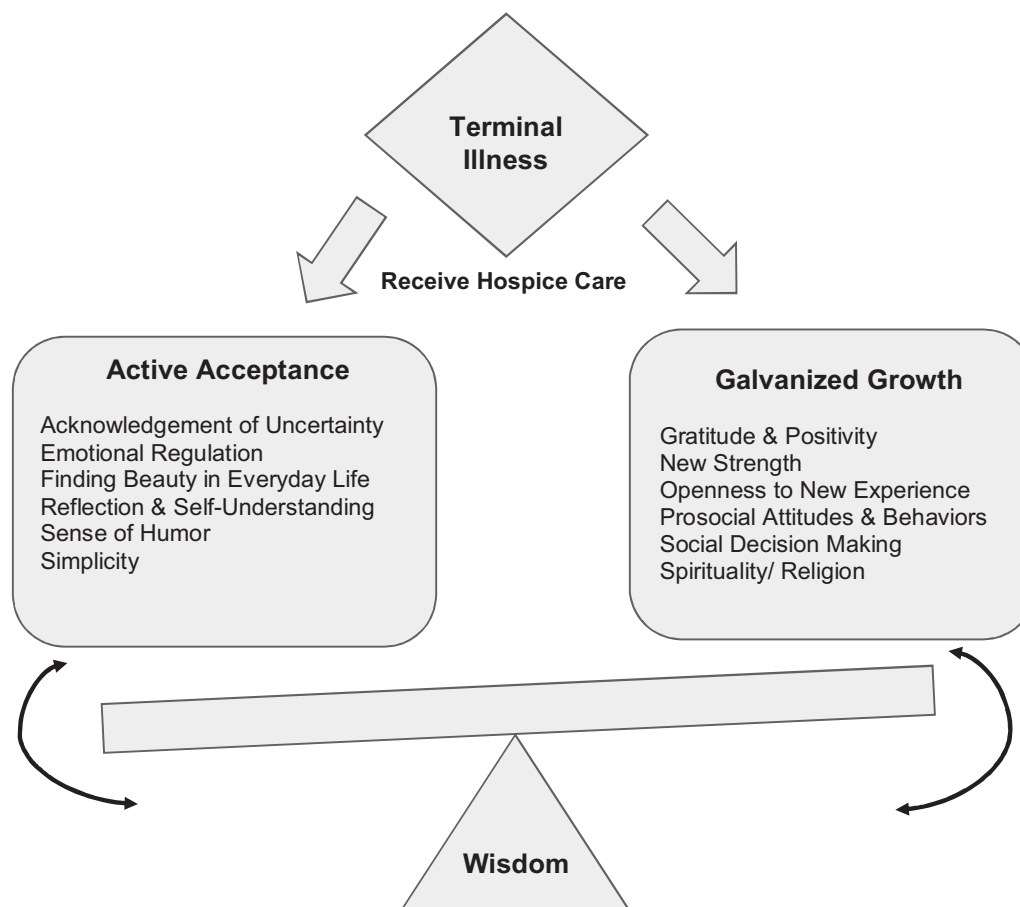


Figure 1. (Colour online) Conceptual model of wisdom as qualitatively described by patients receiving hospice care at the end of their life ($n = 21$).

hospice care changed their overall perspective, how they strived to actively accept their new situation, and how they found a refined sense of galvanized growth in the process. These patients also shed new light on how wisdom involved a delicate balance between learning to simply be, while continually striving to change. Hospice staff may be familiar with how patients seem to “seesaw” in this regard. On some days, patients may appear very accepting of their life circumstances, while on others they seem fraught with struggle. This same process may be mirrored in the actions of patients’ loved ones, and may even be seen among staff as they aim to actively accept patients’ mortality, yet simultaneously work hard to improve the lives of the patients and their families on a daily basis (Meier *et al.*, 2016).

In some respects, perhaps this “seesaw” concept is similar to the balance described in the Serenity Prayer: “Lord, grant me the serenity to accept the things I cannot change, courage to change the things I can, and wisdom to know the difference” (Niebuhr, 2015). In its simplicity, this prayer highlights the dynamic and dualistic nature of

wisdom as described by the hospice patients in this study. They too conveyed a pervasive battle that, when paradoxically embraced, could lead to contentment – even when the worst of life or death is placed before us.

Finally, the hospice patients in this study noted how their struggles related to conscious decisions about the attitudes and behaviors they wished to express in their final days. The patients understood that their final actions would impact their legacy. This finding speaks directly to the importance of not only providing dignity in care, but also ensuring that legacy-related needs of patients are addressed (Chochinov, 2007). In sum, the hospice patients in this study highlighted how, after being diagnosed with a terminal illness, one’s life perspective shifts. One may learn to accept the illness, but may also wisely strive for growth at the same time. This tension can catalyze the need to live each day fully, and by doing so, can leave an even greater legacy.

Although beneficial in understanding perspectives on wisdom among adults in hospice care, this study does have limitations. First, all the participants were drawn from a sample in San

Diego, California, and the majority of patients were Caucasian (81%). Although this percentage mirrors 2014 national statistics with approximately 76% of all hospice patients in the USA being Caucasian (NHPCO Facts and Figures, 2015), it leaves unanswered questions about how a more geographically, ethnically, and racially diverse group of patients might describe wisdom at the end of their lives.

Additionally, because this study required a 60-minute interview, patients with serious cognitive or other psychiatric diagnoses (e.g. major depressive disorder) were not likely to participate. As a result, the components of wisdom found in this study such as active coping and galvanized growth need to be further evaluated within other clinical populations. Finally, the one-time qualitative interview was conducted among patients who were receiving hospice care and, by definition, were in the last six months of life. As a result, all the patients are now deceased, thereby barring any follow-up assessments.

Overall, these study findings suggest that interviewing hospice patients might hold value for understanding not only the concept of wisdom, but other important concepts as well (Morley, 2004). For example, hospice patients may help us refine other conceptualizations about aging, illness, love, loss, or even grace in the face of death (Depp and Jeste, 2006; Montross-Thomas, 2015). Perhaps those living in the last six months of their life could provide an unparalleled and exquisite window into what it means to truly “be alive.”

“In the last analysis, it is our conception of death which decides our answers to all the questions life puts to us.”

— Dag Hammarskjöld, former Secretary-General of the United Nations (1964).

Conflict of interest

The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this report.

Description of authors' roles

Lori P. Montross-Thomas designed and implemented the study, analyzed results, and helped prepare the manuscript. Jamie Joseph and Emily C. Edmonds analyzed data and helped prepare the manuscript. Lawrence A. Palinkas provided qualitative methodology supervision and support in data analysis. Dilip V. Jeste helped design the study,

analyzed data, provided research supervision and support, and edited the manuscript.

Acknowledgments

This study would not have been possible without the courage and commitment of the hospice patients who elected to share their thoughts and reflections as they approached the end of their lives. To them, we are inherently and respectfully grateful. We further wish to acknowledge all the dedicated individuals at Mission Hospice and the San Diego Hospice who were devoted to this study's purpose, design, and completion. This work was supported, in part, by the National Institutes of Health, Grant R25 MH071544/MH/NIMH, the NIMH-funded T32 Geriatric Mental Health Program MH019934, and the Sam and Rose Stein Institute for Research on Aging at University of California San Diego (PI: Dilip V. Jeste) Additional staff time and support were provided by the American Cancer Society 124667-MRSG-13-233-01-PCSM, the Westreich Foundation, and the MAPI Research Trust (PI: Lori P. Montross-Thomas).

References

- Ardelt, M.** (2004). Wisdom as expert knowledge system: a critical review of a contemporary operationalization of an ancient concept. *Human Development*, 47, 257–285. doi:10.1159/000079154.
- Baltes, P. B. and Smith, J.** (2008). The fascination of wisdom: its nature, ontogeny, and function. *Perspectives on Psychological Science*, 3, 56–64. doi:10.1111/j.1745-6916.2008.00062.x.
- Bangen, K. J., Meeks, T. W. and Jeste, D. V.** (2013). Defining and assessing wisdom: a review of the literature. *American Journal of Geriatric Psychiatry*, 21, 1254–1266. doi:10.1016/j.jagp.2012.11.020.
- Bellizzi, K. M.** (2004). Expressions of generativity and posttraumatic growth in adult cancer survivors. *The International Journal of Aging and Human Development*, 58, 267–287. doi:10.2190/DC07-CPVW-4UVE-5GK0.
- Boyatzis, R. E.** (1998). *Transforming Qualitative Information: Thematic Analysis and Code Development*. London; New Delhi: Sage Publications.
- Carstensen, L. L., Isaacowitz, D. and Charles, S. T.** (1999). Taking time seriously: a theory of socioemotional selectivity. *American Psychologist*, 54, 165–181. PMID:10199217.
- Chochinov, H. M.** (2007). Dignity and the essence of medicine: the A, B, C, and D of dignity conserving care. *BMJ*, 334, 184–197. doi:10.1136/bmj.39244.650926.47.
- Clayton, V.** (1975). Erikson's theory of human development as it applies to the aged: wisdom as contradictory cognition. *Human Development*, 18, 119–128.
- Damon, W.** (2000). Setting the stage for the development of wisdom: self-understanding and moral identity during adolescence. In *Understanding Wisdom: Sources, Science, and*

- Society* (pp. 339–360). Philadelphia: Templeton Foundation Press.
- Dedoose** (2015). *Web Application for Managing, Analyzing, and Presenting Qualitative and Mixed Method Research Data*. Los Angeles, CA: SocioCultural Research Consultants, LLC.
- Depp, C. A. and Jeste, D. V.** (2006). Definitions and predictors of successful aging: a comprehensive review of larger quantitative studies. *The American Journal of Geriatric Psychiatry*, 14, 6–20. doi:10.1097/01.JGP.0000192501.03069.bc.
- Diamond, L. M. and Aspinwall, L. G.** (2003). Emotion regulation across the life span: an integrative perspective emphasizing self-regulation, positive affect, and dyadic processes. *Motivation and Emotion*, 27, 125–156.
- Erikson, E. H.** (1950). *Childhood and Society*, revised 1963. New York, NY: Norton.
- Glaser, B. G. and Strauss, A. L.** (1967). *The Discovery of Grounded Theory: Strategies for Qualitative Research*. Chicago: Aldine Publishing Company.
- Glück, J., Bluck, S., Baron, J. and McAdams, D. P.** (2005). The wisdom of experience: autobiographical narratives across adulthood. *International Journal of Behavioral Development*, 29, 197–208. doi:10.1177/01650250444000504.
- Hammar skjöld, D., Auden, W. H. and Sjöberg, L.** (1964). *Markings ["Vägmärken"] by Dag Hammar skjöld. Translated by Leif Sjöberg and W.H. Auden, with a Foreword by WH Auden*. New York, NY: Ballantine Books.
- Happé, F. G. E., Winner, E. and Brownell, H.** (1998). The getting of wisdom: theory of mind in old age. *Developmental Psychology*, 34, 358–362. doi:10.1037/0012-1649.34.2.358.
- Jeste, D. V. and Vahia, I. V.** (2008). Comparison of the conceptualization of wisdom in ancient Indian literature with modern views: focus on the Bhagavad Gita. *Psychiatry*, 71, 197–209. doi:10.1521/psyc.2008.71.3.197.
- Jeste, D. V., Ardel, M., Blazer, D., Kraemer, H. C., Vaillant, G. and Meeks, T. W.** (2010). Expert consensus on characteristics of wisdom: a delphi method study. *The Gerontologist*, 50, 668–680. doi:10.1093/geront/gnq022.
- Kierkegaard, S.** (1843). Journalen JJ. In *Søren Kierkegaard's Skrifter* (vol. 18, p. 306). Copenhagen: Søren Kierkegaard Research Center.
- Kross, E. and Grossmann, I.** (2012). Boosting wisdom: distance from the self enhances wise reasoning, attitudes, and behavior. *Journal of Experimental Psychology: General*, 141, 43–48.
- Lerner, R. M.** (2008). Spirituality, positive purpose, wisdom, and positive development in adolescence: comments on Oman, Flinders, and Thoresen's ideas about "Integrating spiritual modeling into education". *The International Journal for the Psychology of Religion*, 18, 108–118.
- Meier, E., Gallegos, J. V., Thomas, L.P., Depp, C.A., Irwin, S. A. and Jeste, D. V.** (2016). Defining a good death (successful dying): literature review and a call for research and public dialogue. *American Journal of Geriatric Psychiatry*, 24, 261–271.
- Montross-Thomas, L. P.** (2015). What are the most loving moments of your life? *Journal of Palliative Medicine*, 18, 398. doi:10.1089/jpm.2015.0006.18.5.
- Morley, J. E.** (2004). The top 10 hot topics in aging. *The Journals of Gerontology Series A: Biological Sciences and Medical Sciences*, 59, M24–M33. doi:10.1093/gerona/59.1.M24.
- Morse, J. M.** (1995). The significance of saturation. *Qualitative Health Research*, 5, 147–149. doi:10.1177/104973239500500201.
- Murray, S. A., Kendall, M., Grant, E., Boyd, K., Barclay, S. and Sheikh, A.** (2007). Patterns of social, psychological, and spiritual decline toward the end of life in lung cancer and heart failure. *Journal of Pain and Symptom Management*, 34, 393–402. doi:10.1016/j.jpainsymman.2006.12.009.
- NHPCO** (2015). *Facts and Figures: Hospice Care in America*. (vol. 2016, p. 7). Alexandria, VA: National Hospice and Palliative Care Organization.
- Niebuhr, R.** (2015). Major works on religion and politics. In E. Sifton (ed.), *The Library of America*. New York, NY: Literary Classics of the United States, Inc.
- Pasupathi, M., Staudinger, U. M. and Baltes, P.B.** (2001). Seeds of wisdom: adolescents' knowledge and judgment about difficult life problems. *Developmental Psychology*, 37, 351–361. doi:10.1037/0012-1649.37.3.351.
- Pinder, M. M. and Hayslip, B. Jr.** (1981). Cognitive, attitudinal, and affective aspects of death and dying in adulthood: implications for care providers. *Educational Gerontology: An International Quarterly*, 6, 107–123. doi:10.1080/0380127810060201.
- Smith, J. and Baltes, P. B.** (1990). Wisdom-related knowledge: age/cohort differences in response to life-planning problems. *Developmental Psychology*, 26, 494. doi:10.1037/0012-1649.26.3.494.
- Sternberg, R. J.** (1990). *Wisdom: Its Nature, Origins, and Development*. New York, NY: Cambridge University Press.
- Strauss, A. L. and Corbin, J.** (1998). *Basics of Qualitative Research: Techniques and Procedures for Developing Grounded Theory*. Thousand Oaks, CA: Sage.
- Takahashi, M.** (2000). Toward a culturally inclusive understanding of wisdom: historical roots in the East and West. *International Journal of Aging and Human Development*, 51, 217–230. doi:10.2190/H45U-M17W-3AG5-TA49.
- Thomas, M. L. et al.** (2017). A new scale for assessing wisdom based on common domains and a neurobiological model: the San Diego Wisdom Scale (SD-WISE). *Journal of Psychiatric Research*, 1–8. doi:10.1016/j.jpsychires.2017.09.005.
- Vachon, M., Fillion, L. and Achille, M.** (2009). A conceptual analysis of spirituality at the end of life. *Journal of Palliative Medicine*, 12, 53–59. doi:10.1089/jpm.2008.0189.
- Willms, D. G. et al.** (1990). A systematic approach for using qualitative methods in primary prevention research. *Medical Anthropology Quarterly*, 4, 391–409. doi:10.1525/maq.1990.4.4.02a00020.

The relation of close friends to cognitive performance in old age: the mediating role of leisure activities

Andreas Ihle,^{1,2} Michel Oris,² Marie Baeriswyl² and Matthias Kliegel^{1,2}

¹Department of Psychology, University of Geneva, Geneva, Switzerland

²Center for the Interdisciplinary Study of Gerontology and Vulnerability, University of Geneva, Geneva, Switzerland

ABSTRACT

Background: From a conceptual point of view, close friends are an important resource for promoting activity engagement in old age. Leisure activity engagement in turn is a key predictor of cognitive performance. Empirically, it remains unclear so far whether leisure activity engagement mediates between having close friends on the one hand and cognitive performance on the other, which we investigated in a large sample of older adults.

Methods: We assessed cognitive performance (Mill Hill vocabulary scale and Trail Making Test (TMT) parts A and B) in 2,812 older adults. Participants reported information on leisure activity engagement and close friends.

Results: A larger number of leisure activities and a larger number of close friends were significantly related to better cognitive performance in the Mill Hill vocabulary scale and TMT parts A and B. A larger number of close friends were significantly related to a larger number of leisure activities. The number of leisure activities mediated more than half of the relation of the number of close friends to performance in all three cognitive measures.

Conclusions: Having close friends may be helpful to stimulate and promote activity participation in old age. By enhancing individuals' cognitive reserve, this may finally preserve their cognitive performance level in old age.

Key words: cognition, cognitive reserve, leisure activities, close friends, older adults

Introduction

In current gerontological research, a major goal is to understand how interindividual differences in cognitive functioning in old age emerge (Hultsch *et al.*, 1999). To address this issue, the cognitive reserve concept postulates that interindividual differences in the effective recruitment of neural networks and cognitive processes explain differences in individuals' capacity to cope with or compensate for age-related decline (Stern, 2009, 2012). In individuals with healthy cognitive functioning, these mechanisms contribute to the adaptation of brain activity when task difficulty level is increased and thereby enhance cognitive performance (Stern, 2012). Empirically, corroborating the predictions

of the cognitive reserve concept, evidence showed that engaging in leisure activities in old age contributes to the build-up of cognitive reserve and is related to better cognitive functioning such as memory and executive functioning in old age (Karp *et al.*, 2006; Engelhardt *et al.*, 2010; Paillard-Borg *et al.*, 2012; Wang *et al.*, 2013; Ihle *et al.*, 2017; see Hertzog *et al.*, 2008; Opdebeeck *et al.*, 2016, for overviews).

Recently, a focus in research on this topic became to investigate the role of cognitive reserve markers (such as engaging in leisure activities) for explaining the relational pathways from other individual difference characteristics to cognitive functioning in old age (Ihle *et al.*, 2016). Such characteristics may also concern social aspects of older adults' life as, for example, the number of their close friends. This seems reasonable since prior empirical studies showed that having a larger number of close friends is related to better cognitive status and reduced cognitive decline in old age (Barnes *et al.*, 2004; Wang *et al.*, 2015; Ho,

Correspondence should be addressed to: Andreas Ihle, Center for the Interdisciplinary Study of Gerontology and Vulnerability, University of Geneva, Boulevard du Pont d'Arve 28, 1205 Geneva, Switzerland. Phone: +41 22 37 98308. E-mail: Andreas.Ihle@unige.ch. Received 25 Jan 2018; revision requested 4 Apr 2018; revised version received 10 Apr 2018; accepted 25 Apr 2018. First published online 1 June 2018.

2016; Kimura *et al.*, 2017). In general, close friends can serve as or provide access to emotional resources (companionship, positive interactions, and emotional support), instrumental resources (financial support and domestic support), and informational resources (exchange of knowledge and key information; Bourdieu, 1985). These resources may, for example, help to stimulate and enhance an individual's activity participation. As empirically revealed, individuals show a greater engagement in a variety of leisure activities if they have a larger number of close friends (Flora and Segrin, 1998; Huxhold *et al.*, 2014; Hamilton *et al.*, 2017).

From a conceptual perspective, combining the interplay of the aforementioned relationships in one overarching framework, one could predict that activity participation mediates between having close friends on the one hand and cognitive performance in old age on the other. Such mechanisms would be in line with more general gerontological models such as activity theory (Havighurst, 1951) and continuity theory (Atchley, 1989), which postulated that older adults would age successfully if they continued engaging in the same activities as in midlife and, when maintenance was no longer possible, found alternative leisure activities. Yet, even if maintaining a variety of activities or engaging in new activities in old age becomes difficult, with the help of resources and support that close friends can provide, engagement in activities will be promoted, which in turn will have positive aftereffects on cognitive functioning.

Hence, it seems an appealing question whether activity participation mediates between having close friends on the one hand and cognitive performance in old age on the other. However, empirically, this question has not been investigated so far. Therefore, with a large sample of older adults, our specific goal was to investigate whether the number of leisure activities mediated the relation of the number of close friends to cognitive performance (i.e. vocabulary, cognitive processing speed, and cognitive flexibility).

Methods

Participants

Data were collected in 2011 and 2012 and come from the Vivre–Leben–Vivere (VLV) survey (Ludwig *et al.*, 2014; Ihle *et al.*, 2015; Oris *et al.*, 2016), which is a part of the research program LIVES on vulnerability processes across the life course. Participants were randomly selected in the cantonal and federal Swiss administrations' records and stratified by age (65–69 years, 70–74, 75–

79, 80–84, 85–89, and 90+), sex, and canton (Bale, Berne, Geneva, Ticino, and Valais). Two thousand eight hundred and twelve participants with cognitive performance assessment served as sample for the present study. Mean age was 77.9 years ($SD = 8.2$, range 65–101). The sample comprised 1,330 women (47.3%) and 1,482 men (52.7%). All participants gave informed consent and the present study included adherence to the declaration of Helsinki and had been approved by the local ethics commission.

Materials

Cognitive performance

Mill Hill. We administered the Mill Hill scale (Deltour, 1993) measuring vocabulary. For each item, participants had to underline the word (which was intermixed with five distractor words) that semantically matched the target word. After one practice item, participants had to complete ten items. The Mill Hill score was the proportion of correctly completed items.

TMT A. We administered the Trail Making Test part A (TMT A; Reitan, 1958) measuring cognitive processing speed. After seven exercise trails (connecting the numbers from 1 to 8), participants had to connect the numbers from 1 to 25 as fast as possible and without error in ascending order. The TMT A completion time was the time in seconds needed to correctly connect the 25 numbers.

TMT B. We administered the Trail Making Test part B (TMT B; Reitan, 1958) measuring cognitive flexibility. After seven exercise trails (connecting 1-A-2-B-3-C-4-D), participants had to connect the numbers 1–13 in ascending order and the letters A–L in alphabetic order while alternating between numbers and letters (i.e. 1-A-2-B-3-C ... 12-L-13) as fast as possible and without error. The TMT B completion time was the time in seconds needed to correctly connect the 25 numbers/letters.

Number of leisure activities

We interviewed participants regarding their leisure activities such as sports and other physical exercises, go to the cinema, go to conferences, go into a restaurant, journeys, artistic activities, table games, and municipality activities. For analyses, we calculated the overall number of leisure activities participants engaged in.

Number of close friends

We asked participants to indicate the number of their close friends (Barnes *et al.*, 2004; Ho, 2016).

Covariates

We additionally repeated mediation analyses simultaneously controlling for the following covariates: age, sex, highest educational level attained (i.e. primary school level, inferior secondary school level, apprenticeship graduation, superior secondary school level, technical college or superior vocational college degree, or university degree), marital status (i.e. single, married, separated, divorced, or widowed), retirement status (i.e. retired or not), subjective health status (i.e. based on a rating scale ranging from 0 = “worst imaginable health” to 100 = “best imaginable health”), and BMI (i.e. calculated as the self-reported weight in kg divided by the squared self-reported height in m).

Procedure

A face-to-face questionnaire was administered using the CAPI (Computer Assisted Personal Interview) method. This session contained (besides a larger set of other questionnaires) a socio-demographic survey, the questions regarding leisure activities and close friends, and the paper-pencil assessment of the three cognitive measures. Participants were individually tested by trained research personnel, who always assured that participants fully understood and followed the instructions.

Statistical analyses

The following analyses were run: We inspected relations between performance in Mill Hill, TMT A, TMT B, the number of leisure activities, and the number of close friends (by calculating Pearson's correlation coefficients r). Regarding our specific goal, we investigated whether the relation of the number of close friends to performance in Mill Hill/TMT A/TMT B was mediated via the number of leisure activities. For these mediation analyses, we used a path model approach, with an individual mediation model for each cognitive measure. These models contained three paths (see Figure 1 for an illustration): path a, the number of leisure activities regressed on the number of close friends; path b, performance in Mill Hill/TMT A/TMT B regressed on the number of leisure activities; and path c, performance in Mill Hill/TMT A/TMT B regressed on the number of close friends. Importantly, for evaluating mediation, the applied path model approach allowed simultaneously estimating the residual direct (non-mediated) relation of the number of close friends to performance in Mill Hill/TMT

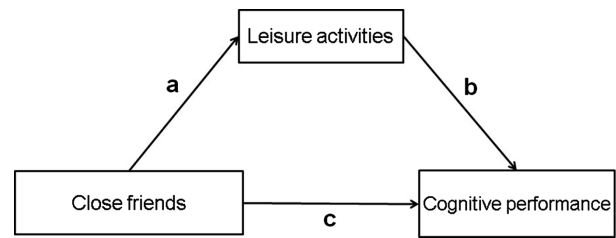


Figure 1. General structure of the path models applied to investigate whether the relation of the number of close friends to cognitive performance in old age was mediated via the number of leisure activities. These models allowed simultaneously estimating the residual direct (non-mediated) relation of the number of close friends to performance in Mill Hill/TMT A/TMT B (c) and the indirect (mediated) relation via the number of leisure activities ($a \cdot b$).

A/TMT B (i.e. the coefficient of path c) and the indirect (mediated) relation via the number of leisure activities (i.e. the product of the coefficients for paths a and b), including their significance. The proportion of the size of the mediated relation and the total relation (i.e. the sum of the mediated and the non-mediated direct relation) allowed quantifying the portion of the relation of the number of close friends to performance in Mill Hill/TMT A/TMT B that was exerted indirectly via the number of leisure activities as mediator. For all analyses, to achieve that higher values represented better performance across all variables (as common in correlative studies), for TMT A and TMT B, the distribution of completion time of all participants was reversed based on the sample mean so that interindividual differences remained identical.

Results

Descriptive statistics

Mean performance in Mill Hill was 59.4% correct ($SD = 25.7$). Mean completion time in TMT A was 66.20 sec ($SD = 30.61$). Mean completion time in TMT B was 127.59 sec ($SD = 52.66$). Mean number of leisure activities was 8.15 ($SD = 3.50$). Mean number of close friends was 1.90 ($SD = 1.23$).

Bivariate relations between measures

A larger number of leisure activities and a larger number of close friends were significantly related to better performance in all three cognitive measures. In addition, a larger number of close friends were significantly related to a larger number of leisure activities (see Table 1 for the full descriptive correlation matrix).

Table 1. Full descriptive correlation matrix of measures

| VARIABLE | 1 | 2 | 3 | 4 | 5 |
|---------------------------------|---------|---------|---------|---------|---|
| 1. Mill Hill | – | | | | |
| 2. TMT A | 0.24*** | – | | | |
| 3. TMT B | 0.28*** | 0.62*** | – | | |
| 4. Number of leisure activities | 0.23*** | 0.39*** | 0.33*** | – | |
| 5. Number of close friends | 0.11*** | 0.17*** | 0.12*** | 0.27*** | – |

*** $p < 0.001$.

Table 2. Results of mediation analyses

| VARIABLE | INDIRECT RELATION | RESIDUAL DIRECT RELATION |
|-----------|-------------------|--------------------------|
| Mill Hill | 0.06*** (50.5%) | 0.05** |
| TMT A | 0.10*** (59.1%) | 0.07** |
| TMT B | 0.07*** (58.2%) | 0.05* |

Note. Results of mediation analyses to investigate whether the relation of the number of close friends to performance in Mill Hill/TMT A/TMT B was mediated via the number of leisure activities. Left panel: Values represent indirect (mediated) relation sizes β . In parentheses, the portion of the relation of the number of close friends to performance in Mill Hill/TMT A/TMT B that was exerted indirectly via the number of leisure activities is given. Right panel: Values represent residual direct (non-mediated) relation sizes β .

*** $p < 0.001$.

** $p < 0.01$.

* $p < 0.05$.

Mediation analyses

Regarding our specific goal, we found that the relation of the number of close friends to all three cognitive measures was partly mediated via the number of leisure activities. Specifically, the number of leisure activities mediated about 50.5%–59.1% of the relation of the number of close friends to cognitive performance (see Table 2 for an overview).

Additional control analyses

We additionally repeated mediation analyses simultaneously controlling for age, sex, educational level, marital status, retirement status, subjective health status, and BMI. In these control analyses, the mediation of the relation of the number of close friends to all three cognitive measures via the number of leisure activities remained significant. The number of leisure activities mediated about 53.0%–85.6% of the relation of the number of close friends to cognitive performance (see Table 3 for an overview).

Table 3. Results of mediation analyses (controlling for covariates)

| VARIABLE | INDIRECT RELATION | RESIDUAL DIRECT RELATION |
|-----------|-------------------|--------------------------|
| Mill Hill | 0.03*** (85.6%) | <0.01 ns |
| TMT A | 0.06*** (53.0%) | 0.05** |
| TMT B | 0.04*** (69.7%) | 0.02 ns |

Note. Results of mediation analyses to investigate whether the relation of the number of close friends to performance in Mill Hill/TMT A/TMT B was mediated via the number of leisure activities, simultaneously controlling for age, sex, educational level, marital status, retirement status, subjective health status, and BMI. Left panel: Values represent indirect (mediated) relation sizes β . In parentheses, the portion of the relation of the number of close friends to performance in Mill Hill/TMT A/TMT B that was exerted indirectly via the number of leisure activities is given. Right panel: Values represent residual direct (non-mediated) relation sizes β .

*** $p < 0.001$.

** $p < 0.01$.

ns = non-significant, $p > 0.05$.

Discussion

The present study set out to investigate whether the number of leisure activities mediated the relation of the number of close friends to cognitive performance in a large sample of older adults. First of all, results showed that greater engaging in leisure activities was related to better performance in all three investigated cognitive measures. This further corroborates the conceptual view that an active lifestyle in late adulthood may further contribute to cognitive reserve, thereby being related to better cognitive status in old age (Hertzog *et al.*, 2008; Ihle *et al.*, 2015; Opdebeeck *et al.*, 2016). Furthermore, again coherently for all three investigated cognitive measures, our results are in line with prior empirical studies showing that having a larger number of close friends is related to better cognitive performance in old age (Barnes *et al.*, 2004; Wang *et al.*, 2015; Ho, 2016; Kimura *et al.*, 2017). Present results also confirm that individuals show a greater engagement in a variety of leisure activities if they have a larger number of close friends (Flora and Segrin, 1998; Huxhold *et al.*, 2014; Hamilton *et al.*, 2017).

Following our specific goal, notably coherently for all three investigated cognitive measures, we found that activity participation mediated between having close friends on the one hand and cognitive performance in old age on the other. Specifically, the number of leisure activities mediated more than half of the relation of the number of close friends to cognitive performance. Importantly, this finding was robust to adjustment for a wide array of potentially confounding variables including age, sex, educational level, marital status, retirement status, subjective health status, and BMI. This

confirms that close friends are an important resource in old age (Bourdieu, 1985; Barnes *et al.*, 2004; Huxhold *et al.*, 2014; Wang *et al.*, 2015; Kimura *et al.*, 2017). For instance, if maintaining a variety of activities or engaging in new activities in old age becomes difficult, with the help of resources and support that close friends can provide, engagement in activities could be promoted, which in turn may have positive aftereffects on cognitive functioning. This also suggests that for cognitive functioning in old age it seems not only important to have a number of close friends, but also to use the resources and support offered by those friends such as encouragements to pursue an active life. In conclusion, present results suggest that having close friends may be helpful to stimulate and promote activity participation in old age. By enhancing individuals' cognitive reserve, this may finally preserve their cognitive performance level in old age.

In terms of possible practical and policy implications, one could argue that promoting activity participation with the help of older adults' social networks might perhaps contribute to their cognitive health. However, given the limitations of present cross-sectional data, we underline that future longitudinal studies will have to further investigate the present observations before making suggestions. In the context of the latter notions, we acknowledge that the present study is limited by its cross-sectional design that does not allow for causal inferences. The directionality of observed relationships cannot be established with the present cross-sectional data. Thus, we cannot clearly disentangle between individuals who decline because of reducing activities and those who reduce activities because of cognitive decline. More generally, analyses of the present study give only information about interindividual differences in cognitive status but do not allow drawing conclusions regarding cognitive decline (i.e. intraindividual changes over time). For instance, present observations may be affected by selection issues. Those individuals with lower cognitive functioning from early in life may attract fewer friends and may pursue fewer leisure activities later in life. Moreover, although present findings were robust to adjustment for a wide array of potentially confounding variables including age, sex, educational level, marital status, retirement status, subjective health status, and BMI, there may be other covariates that have not been assessed and therefore could not be controlled for. Thus, the present study identifies important associations, but cannot address causal relations. Therefore, the present study may stimulate future longitudinal research assessing changes in a comprehensive set of cognitive domains, activities, and social networks

to gain further insights into the interplay of having close friends, engagement in leisure activities, and cognitive functioning in old age.

Conflict of interest

None.

Description of authors' roles

A. Ihle formulated the research question, analyzed the data, and wrote the manuscript. M. Oris and M. Baeriswyl formulated the research question, conceptualized the study, supervised the data collection, and participated in writing. M. Kliegel contributed to formulate the research question and was involved in writing the manuscript.

Acknowledgments

All four authors belong to the Swiss National Centre of Competence in Research LIVES – Overcoming vulnerability: life course perspectives, which is financed by the Swiss National Science Foundation (grant number: 51NF40-160590). The authors are grateful to the Swiss National Science Foundation for its financial assistance.

References

- Atchley, R. C. (1989). A continuity theory of normal aging. *Gerontologist*, 29, 183–190.
- Barnes, L. L., de Leon, C. F. M., Wilson, R. S., Bienias, J. L. and Evans, D. A. (2004). Social resources and cognitive decline in a population of older African Americans and whites. *Neurology*, 63, 2322–2326.
- Bourdieu, P. (1985). The forms of capital. In J. G. Richardson (ed.), *Handbook of Theory and Research for the Sociology of Education* (pp. 241–258). New York, NY: Greenwood Press.
- Deltour, J. (1993). *Echelle de Vocabulaire Mill Hill de J.C. Raven*. [Raven Mill Hill Vocabulary Scale]. Braine-le Chateau, Belgium: Editions l'Application des Techniques Modernes.
- Engelhardt, H., Buber, I., Skirbekk, V. and Prskawetz, A. (2010). Social involvement, behavioural risks and cognitive functioning among older people. *Ageing & Society*, 30, 779–809.
- Flora, J. and Segrin, C. (1998). Joint leisure time in friend and romantic relationships: the role of activity type, social skills and positivity. *Journal of Social and Personal Relationships*, 15, 711–718.
- Hamilton, K., Warner, L. M. and Schwarzer, R. (2017). The role of self-efficacy and friend support on adolescent vigorous physical activity. *Health Education & Behavior*, 44, 175–181.

- Havighurst, R. J.** (1951). *Developmental Tasks and Education*. New York, NY: Longmans Green.
- Hertzog, C., Kramer, A. F., Wilson, R. S. and Lindenberger, U.** (2008). Enrichment effects on adult cognitive development: can the functional capacity of older adults be preserved and enhanced? *Psychological Science in the Public Interest*, 9, 1–65.
- Ho, C. Y.** (2016). The relationship from friendship links to educational achievement. *B. E. Journal of Economic Analysis & Policy*, 16, 1563–1572.
- Hultsch, D. F., Hertzog, C., Small, B. J. and Dixon, R. A.** (1999). Use it or lose it: engaged lifestyle as a buffer of cognitive decline in aging? *Psychology and Aging*, 14, 245–263.
- Huxhold, O., Miche, M. and Schuz, B.** (2014). Benefits of having friends in older ages: differential effects of informal social activities on well-being in middle-aged and older adults. *Journals of Gerontology Series B: Psychological Sciences and Social Sciences*, 69, 366–375.
- Ihle, A., Oris, M., Fagot, D., Maggiori, C. and Kliegel, M.** (2016). The association of educational attainment, cognitive level of job, and leisure activities during the course of adulthood with cognitive performance in old age: the role of openness to experience. *International Psychogeriatrics*, 28, 733–740.
- Ihle, A., Oris, M., Fagot, D., Baeriswyl, M., Guichard, E. and Kliegel, M.** (2015). The association of leisure activities in middle adulthood with cognitive performance in old age: the moderating role of educational level. *Gerontology*, 61, 543–550.
- Ihle, A. et al.** (2017). High-Density lipoprotein cholesterol level relates to working memory, immediate and delayed cued recall in Brazilian older adults: the role of cognitive reserve. *Dementia and Geriatric Cognitive Disorders*, 44, 84–91.
- Karp, A., Paillard-Borg, S., Wang, H. X., Silverstein, M., Winblad, B. and Fratiglioni, L.** (2006). Mental, physical and social components in leisure activities equally contribute to decrease dementia risk. *Dementia and Geriatric Cognitive Disorders*, 21, 65–73.
- Kimura, D., Takeda, T., Ohura, T. and Imai, A.** (2017). Evaluation of facilitative factors for preventing cognitive decline: a 3-year cohort study of community intervention. *Psychogeriatrics*, 17, 9–16.
- Ludwig, C., Cavalli, S. and Oris, M.** (2014). “Vivre/Leben/Vivere”: an interdisciplinary survey addressing progress and inequalities of aging over the past 30 years in Switzerland. *Archives of Gerontology and Geriatrics*, 59, 240–248.
- Opdebeeck, C., Martyr, A. and Clare, L.** (2016). Cognitive reserve and cognitive function in healthy older people: a meta-analysis. *Aging, Neuropsychology, and Cognition*, 23, 40–60.
- Oris, M. et al.** (2016). Representation of vulnerability and the elderly. A total survey error perspective on the VLV survey. In M. Oris, C. Roberts, D. Joye and M. Ernst-Stähli (eds.), *Surveying Human Vulnerabilities across the Life Course* (pp. 27–64). Heidelberg: Springer.
- Paillard-Borg, S., Fratiglioni, L., Xu, W. L., Winblad, B. and Wang, H. X.** (2012). An active lifestyle postpones dementia onset by more than one year in very old adults. *Journal of Alzheimers Disease*, 31, 835–842.
- Reitan, R. M.** (1958). Validity of the trail making test as an indicator of organic brain damage. *Perceptual and Motor Skills*, 8, 271–276.
- Stern, Y.** (2009). Cognitive reserve. *Neuropsychologia*, 47, 2015–2028.
- Stern, Y.** (2012). Cognitive reserve in ageing and Alzheimer’s disease. *Lancet Neurology*, 11, 1006–1012.
- Wang, B. Y., He, P. and Dong, B. R.** (2015). Associations between social networks, social contacts, and cognitive function among Chinese nonagenarians/centenarians. *Archives of Gerontology and Geriatrics*, 60, 522–527.
- Wang, H. X. et al.** (2013). Late life leisure activities and risk of cognitive decline. *Journals of Gerontology Series A: Biological Sciences and Medical Sciences*, 68, 205–213.

Spirituality, social support, and flexibility among older adults: a five-year longitudinal study

Nathalie Bailly,¹ Guillaume Martinet,² Claude Ferrand,¹ Océane Agli,¹
Caroline Giraudeau,¹ Kamel Gana³ and Nicolas Roussiau⁴

¹EA 2114, Psychologie des Ages de la Vie et Adaptation, University of Tours, Tours, France

²Laboratory of Vulnerability and Innovations in Sport, EA 7428, University of Claude Bernard Lyon 1 – Univ Lyon, Lyon, France

³EA 4139, Santé et Qualité de Vie, University of Bordeaux, Bordeaux, France

⁴Laboratory LPPL, University of Nantes, Nantes, France

ABSTRACT

Objectives: The objectives of the study were to examine the trajectory of spirituality among older adults, to investigate the roles of gender and religion on the developmental trajectory of spirituality, and to explore whether the linear growth of spirituality accelerated or decelerated at time points at which the participants reported high scores of social support and flexibility.

Design: A five-year longitudinal study.

Setting: The research used data from a longitudinal study, which follows a non-institutionalized older adults cohort of residents from France. The data used in this paper were collected at three time points (T1: 2007; T2: 2009; T3: 2012).

Participants: A total of 567 participants were included in the analysis (59.44% female; $M_{age} = 75.90$, $SD = 5.12$).

Measurements: Multilevel growth curve analysis was used measuring spirituality, satisfaction with social support, and flexibility.

Results: The results indicated the following: (1) stability of spirituality over time, (2) older women reported higher levels of spirituality than older men, and those who had a religion reported higher scores of spirituality than their counterparts who had no religion (these effects were strong and clinically meaningful), (3) older adults who reported higher levels of social support and flexibility also reported higher levels of spirituality, and (4) the slope of spirituality seemed to accelerate at time points at which participants also had higher levels of social support and flexibility (these effects were rather small but of theoretical interest).

Conclusion: The results of the present study help to improve the understanding of the potential benefit of encouraging the spiritual aspects of life.

Key words: aging, longitudinal studies, spirituality, religion

Introduction

The spirituality dimension of older adults has recently been integrated in the field of successful aging because of its links with health and well-being outcomes (Tomás *et al.*, 2016). Spirituality has long been associated with religion. Hill and Pargament (2008) defined religiosity as a reference to external, institutionalized, formal, and doctrinal

aspects of religious life, whereas spirituality refers to a personal subjective experience. Spirituality is a search for meaning, involving the questioning of life and the relationship with the sacred or transcendent, which may or may not stem from the development of religious rituals and the formation of a community (Koenig *et al.*, 2001). From a theoretical perspective, Erikson's (1987) psychosocial theory of development and Tornstam's (2005) gerotranscendence theory argue that as age increases, many people perceive themselves as having increasingly transcendent attitudes. They take more delight in their inner world, are less fearful of death, and feel a greater connection to the entire universe. In a review

Correspondence should be addressed to: Nathalie Bailly, Department of Psychology, EA 2114, Psychologie des Ages de la Vie, 3 Rue des Tanneurs, 30041 Tours Cedex 01, France. Phone: +33 2 47 36 65 56. Email: nathalie.bailly@univ-tours.fr. Received 22 Sep 2017; revision requested 5 Nov 2017; revised version received 20 Dec 2017; accepted 21 Dec 2017. First published online 30 January 2018.

of spiritual change with aging, Dalby (2006) indicated an increase in spirituality or aspects of gerotranscendence with age. In a longitudinal study across a life span of over 60 years, Wink and Dillon (2002) suggested that levels of spirituality increased significantly especially from middle to late adulthood, with women evidencing a higher level of spirituality than men. Although some experimental studies have explored changes in levels of spirituality over the course of people's lives (i.e. across the life span), surprisingly a few studies have investigated trajectories of spirituality among older adults. However, spirituality may offer a purpose and meaning toward the end of life and provides a framework for managing concerns and decisions at this time. Thus, the first objective of the present study was to examine the trajectory of spirituality among older adults. Based on previous results, we assumed that there would be an increase in spirituality over time. We also examined the roles of gender and religion on the trajectory of spirituality. We expected higher scores of spirituality for women and for those who report having religious beliefs than for men and for those reporting no religion.

Several mechanisms to explain relationships between spirituality and health or well-being outcomes have been reported (Zimmer *et al.*, 2016). Among them, social support has been raised as a main contributing factor that could be explored further. Social support is an essential element of well-being, especially among older adults (Seeman, 2000). The needs to belong, to trust others, and to rely on each other are fundamental human motivations, which give meaning of life. In particular, perceived social support, which has consistently been associated with various positive outcomes, appears to be more important than received social support (Cohen *et al.*, 2000). On this basis, we can expect a relationship between spirituality and satisfaction with social support.

The importance of maintaining a sense of control over personal development for better psychological well-being has largely been acknowledged as a primordial factor among a wide range of developmental regulation theories. The role of goal adjustment among aging processes has been developed in the dual process model of Brandtstädter (Brandtstädter and Rothermund, 2002). This model highlights that there is an age-related shift toward flexibility over tenacity. According to Brandtstädter, flexibility enhances the disengagement from instrumental striving and the reorientation toward intrinsic, self-transcending goals under declining life-time reserves. By undermining future-related goals, a growing awareness

of life's finitude should also strengthen an orientation toward values and sources of meaning that transcend the horizon of personal lifetime (Brandtstädter *et al.*, 2010). On this basis, we can expect a relationship between spirituality and flexibility.

In sum, based on a longitudinal study, the first objective of the present study was to examine the developmental trajectory of spirituality among older adults. We expected an increase in spirituality over time. In addition, we investigated the roles of gender and religion on the developmental trajectory of spirituality among older adults. We expected that older women as well as religious individuals would have higher scores of spirituality. The second objective was to explore the within-person synergies of spirituality, social support, and flexibility. We hypothesized that trajectories of spirituality were accelerated at time points at which older adults reported high scores of social support and flexibility.

Methods

Participants and procedure

This research used data from an ongoing longitudinal study initiated in 2001 (for more details, see Alaphilippe *et al.*, 2005) by a team of researchers at the University of Tours (France), and which follows a non-institutionalized older adults cohort of residents from France (recruited through an advertisement in a specialized journal). The survey was mailed and participants returned the completed questionnaire in a prepaid envelop. Anonymity was ensured by attributing an identification number to each participant. Data were collected every two years between 2001 and 2012. On the first assessment in 2001, the sample of this cohort comprised 906 participants with a mean age of 72.5 years ($SD = 5.89$, range = 62–95). In 2001, 2003, and 2005, spirituality was not assessed. Thus, the data used in this paper were collected at three time points, with participants participating in up to three waves of measurement (T1: 2007; T2: 2009; T3: 2012). Data for spirituality were available in 2007 (T1) for 567 participants (48.15% married or had a partner; 59.44% female; $M_{age} = 75.90$, $SD = 5.12$), in 2009 (T2) for 387 participants (51.68% married or had a partner; 58.91% female; $M_{age} = 79.23$, $SD = 4.96$), and in 2012 (T3) for 362 participants (49.45% married or had a partner; 60.22% female; $M_{age} = 81.80$, $SD = 4.65$). The sample included a large proportion of participants (i.e. 78.3%) who reported having a religion with older women reporting more religious beliefs than older men ($\chi^2(1) = 6.87$, $p < 0.001$).

Table 1. Sample characteristics and descriptive statistics

| | 2007 N = 567 | 2009 N = 387 | 2012 N = 362 |
|--|-----------------|-----------------|-----------------|
| Age, <i>M</i> (<i>SD</i>) | 75.9 (5.1) | 79.2 (4.9) | 81.8 (4.7) |
| Male, % (<i>n</i>) | 40.6% (230) | 41.1% (159) | 39.8% (144) |
| Religious persons, % (<i>n</i>) | 78.3% (N = 444) | 78.3% (303) | 78.5% (284) |
| Marital status, % (<i>n</i>) | | | |
| With partner | 48.1% (273) | 51.7% (200) | 49.4% (179) |
| Without partner | 51.9% (295) | 48.3% (187) | 50.6% (183) |
| Years of education, <i>M</i> (<i>SD</i>) | 10.2 (2.5) | 10.4 (2.4) | 10.5 (2.6) |
| Financial satisfaction ^a , <i>M</i> (<i>SD</i>) [1–4] | 3 (0.7) | 3 (0.6) | 3 (0.6) |
| Health evaluation ^b , <i>M</i> (<i>SD</i>) [1–5] | 3 (0.6) | 2.9 (0.5) | 2.9 (0.6) |
| Number of diseases ^c , <i>M</i> (<i>SD</i>) | 2.4 (1.5) | 2.5 (1.7) | 2.5 (1.7) |
| Spirituality, <i>M</i> (<i>SD</i>) [1–6] | 4.5 (1.1) | 4.4 (0.7) | 4.3 (1.1) |
| Flexible goal adjustment, <i>M</i> (<i>SD</i>) [1–5] | 3.7 (0.5) | 3.7 (0.5) | 3.7 (0.5) |
| Social support satisfaction, <i>M</i> (<i>SD</i>) [0–28] | 2.9 (0.6) | 3 (0.5) | 3 (0.6) |

M = Mean, *SD* (standard deviation), [min–max].

^aFinancial satisfaction was assessed with the question: “How would you describe your present financial situations: Very dissatisfied (1), Dissatisfied (2), Satisfied (3), Very satisfied (4).”

^bHealth evaluation was assessed by the question: “In general, would you say your health is very poor (1), poor (2), good (3), very good (4), or excellent (5)?”

^cThe number of diseases was evaluated with the Multidimensional Functional Assessment Questionnaire. This scale uses a list of 26 common diseases in older persons (such as diabetes, chronic bronchitis, hypertension, arthritis, gastrointestinal diseases, cardiovascular diseases, and cancer). The number of illnesses was measured by the total number of diseases reported by each participant.

Among those with a religion, there were 92.74% Catholics, 6.18% Protestants, 0.81% Buddhists, and 0.27% Jews. (Religion was assessed by the question: “Which is your religious affiliation? For further analyses, religion was coded as a dummy variable with religious persons (1) or not religious persons (2).)

Across the three waves, the participants were satisfied with their financial situation and considered their health as “good.” They had a few diseases. The most frequent diseases encountered over the five years were arthritis (57%), hypertension (27.5%), gastrointestinal diseases (22.3%), and cardiovascular diseases (18%). The main sample characteristics are presented in Table 1.

Refusal, low cognitive performance, and death are the common reasons for attrition in prospective studies of older adults. In the present study, retention rates were 68% between the first and second waves and 93% between the second and third waves. To investigate the potential impact of attrition, differences in age, spirituality, social support, and flexibility were tested between participants who completed the measures during the three waves and those who dropped out of the study before Time 3. Although their scores of spirituality and flexibility did not differ, participants who dropped out were older ($p < 0.001$) and reported lower levels social support at T1

($p = 0.004$) than those who completed the three waves.

Measures

Spirituality was assessed using a French version of Underwood and Teresi’s (2002) daily spiritual experience scale (DSES; Bailly and Roussiau, 2010). This short scale has been validated in an older population and contains six items. It is intended to assess a person’s perception of the transcendent (God, the divine) in daily life. The six items are rated using a Likert scale, in which the response categories are “many times a day,” (6) “every day,” (5) “most days,” (4) “some days,” (3) “once in a while,” (2) and “never or almost never” (1). High scores on DSES indicate high spirituality. In the present sample, Cronbach’s α ’s of the DSES were 0.90 at T1, 0.92 at T2, and 0.91 at T3.

Social support was assessed using the subscale satisfaction with social support of the Duke social support index (Koenig *et al.*, 1993). This scale contains seven items. Items are rated on a 5-point Likert scale, ranging from “never” to “most of time.” Higher scores indicate more satisfaction with social support. In the present sample, Cronbach’s α ’s of the social support were 0.81 at T1, 0.82 at T2, and 0.83 at T3.

Flexible goal adjustment (FGA) was assessed using a French version of Brandtstädter and

Renner's (1990) FGA scale. FGA contains ten items. Items are rated on a 5-point Likert scale, ranging from 1 "strongly disagree" to 5 "strongly agree." High scores on the FGA scale indicate high accommodative flexibility. In the present sample, Cronbach's α 's of the FGA were 0.68 at T1, 0.74 at T2, and 0.74 at T3.

Data analysis

Multilevel growth curve analyses (MGCA) were used to examine the trajectory of spirituality in older adults (Singer and Willett, 2003). Considering that MGCA allows the number of available measurement waves to differ across individuals, all participants that completed the questionnaire of spirituality at least once during the study could be incorporated in the model (Singer and Willett, 2003). All analyses were conducted using the R package labeled *lme4* (using linear function with p-values estimated based on Satterthwaite's approximations) using the restricted maximum likelihood algorithm for handling missing data (Bates *et al.*, 2015).

First, a two-level model estimated both the average growth and the individual differences in growth in order to examine the slope of spirituality over time. At Level 1, age (linear trajectory) was entered as a continuous predictor to estimate the average intercept (β_0) and the average linear growth (β_1) of the continuous outcome variable of interest (spirituality). We also tested a model in which age (linear trajectory) and squared age (quadratic trajectory) were entered as continuous predictors to estimate the average intercept (β_0), the average linear growth (β_1), and the average quadratic growth (β_2). However, the quadratic slope did not improve the model fit and was not included in further analyses. The age variable was centered at the initial age of the participants. Thus, the intercept should be interpreted as the level of spirituality experienced at 66 years. The random effect of both the intercept and the linear slope was included in each of the models.

Second, a MGCA model estimated the effect of gender and religion (Level 2 ordinal predictors) on the initial level of the continuous outcome variable spirituality (intercept as outcome model) as well as the cross-level moderating roles of gender and religion in the linear developmental trajectory of the continuous outcome variable spirituality (i.e. slope as outcome model). In all analyses, gender and religion were dummy coded (1 = male or no religion; 2 = female or religion). Third, a MGCA model was designed to test, in addition to the aforementioned effects of gender and religion, whether the linear growth of spirituality accelerated

or decelerated at time points at which the scores of the participants on social support and flexibility were higher than their own average (within-person interactions between age and the other variables). Therefore, this model estimated (in addition to the sex and religion effects previously specified in the second model) the within-person (as Level 1 continuous predictors (flexibility level 1 and social support level 1) with group mean centering) and the between-person (as Level 2 continuous predictors (flexibility level 2 and social support level 2) with grand mean centering) main effects of both social support and flexibility on the continuous outcome variable (i.e. spirituality). Moreover, the within-level moderating effects of social support and flexibility (Level 1 continuous predictors social support \times age and flexibility \times age) in the developmental trajectory of the continuous outcome variable of spirituality were also included into the third model.

Results

Development of spirituality in older adults (Model 1)

The results of the MGCA model revealed a non-significant linear effect of age ($b = -0.03$, $p > 0.10$) on spirituality (see Model 1 in Table 2).

Effect of gender and religion on spirituality in older adults (Model 2)

Gender and religion were significantly associated with spirituality ($b = 1.97$ and 5.24 , $p < 0.05$). In other words, older women reported a higher level of spirituality (i.e. intercept) than older men. Similarly, older adults who had religious beliefs reported a higher level of spirituality than those who had no religion. Moreover, gender did not significantly moderate the developmental trajectory of spirituality over time ($b = -0.03$, $p > 0.10$). This result means that spirituality was significantly higher for older women than older men and that there is no evidence for a differential effect of gender across age. Religion does not significantly moderate the linear developmental trajectory of spirituality over time ($b = 0.13$, $p = 0.07$) (see Model 2 in Table 2).

Within-person synergies of spirituality, social support, and flexibility (Model 3)

At Level 2 (between-person main effects of social support and flexibility on spirituality), social support ($b = 0.27$, $p < 0.05$) and flexibility ($b = 0.35$, $p < 0.05$) were significantly associated with spirituality (see Model 3 in Table 2). In other

Table 2. Unstandardized parameter estimates of the spirituality growth curve models

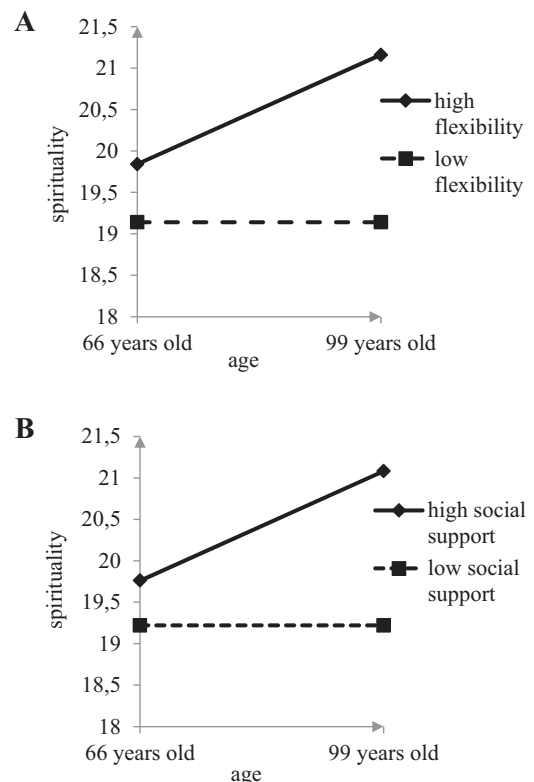
| | MODEL 1 | MODEL 2 | MODEL 3 |
|--|---------------|----------------------------|----------------------------|
| Fixed effects – Estimates (standard errors) | | | |
| Intercept | 19.15*(0.44) | 19.43 (0.78*) | 19.49 (0.76)* |
| Age | – 0.03 (0.03) | 0.01 (0.05) | 0.02 (0.05) |
| Gender | | 1.97 (0.93)* | 1.94 (0.90)* |
| Gender × age | | – 0.03 (0.06) | – 0.05 (0.05) |
| Religion | | – 5.24 (1.12)* | – 5.10 (1.08)* |
| Religion × age | | – 0.13 (0.07) [†] | – 0.12 (0.07) [†] |
| Social support | | | – 0.11 (0.13) |
| Flexibility | | | – 0.13 (0.09) |
| Social support × age | | | 0.02 (0.01)* |
| Flexibility × age | | | 0.02 (0.01)* |
| Social support level 2 | | | 0.27 (0.09)* |
| Flexibility level 2 | | | 0.35 (0.08)* |
| Random effects – Variance (standard deviation) | | | |
| Intercept | 48.41 (6.96) | 42.47 (6.52) | 36.64 (6.05) |
| Age | 0.04 (0.20) | 0.04 (0.19) | 0.04 (0.20) |
| Residual | 6.09 (2.47) | 6.16 (2.48) | 5.98 (2.45) |
| –2 × log likelihood | 7330.9 | 6642.8 | 6453.4 |

* $p < 0.05$.[†] $p \leq 0.08$.

words, older adults who reported higher levels of social support and flexibility also reported higher levels of spirituality (intercept). Otherwise, the within-person interactions (i.e. Level 1 predictors) of age with social support ($b = 0.02$, $p < 0.05$) and flexibility ($b = 0.02$, $p < 0.05$) reached significance. Therefore, the linear increase in spirituality seems to have accelerated at time points at which participants also had higher ($b = 0.04$) rather than lower ($b = 0.00$) levels of social support and flexibility (see Figure 1 for a visual depiction of these effects). Alternatively, the significant within-person interactions can be taken as evidence to indicate that the strength of the association of spirituality with social support and flexibility increased over time among older adults.

Discussion

The aim of the present study was to explore the trajectory of spirituality among older adults over a five-year follow-up. We hypothesized that there would be an increase in spirituality over time, but the results from the MGCA revealed a non-significant effect of age on spirituality. In our study, the mean rating score was relatively high (4.4/6). We can assume that our participants had found meaning in their lives and had thus reached a higher level of spirituality. The question remains as to when spirituality begins to increase. One of the rare longitudinal studies conducted on spiritual development (Wink and Dillon, 2002)

**Figure 1.** Depiction of the interaction terms of flexibility × age (panel A) and social support × age (panel B).

indicates an increase in spirituality between late middle adulthood (50–60) and older adulthood (60–70). Other studies that highlighted a rise in

spirituality over time used a cross-sectional design comparing younger and older adults (Tornstam, 2005; Brown *et al.*, 2013) and the age range of our participants (66–95 years), and also the follow-up period (five years) may have been insufficient to investigate the change in spirituality throughout the life-span. In a cross-sectional analysis, Brown *et al.* (2013) indicated a non-linear age effect on the spiritual transcendence scale with spirituality rising over late adolescence and the adult life course and with lower scores for older adulthood than those for middle age. More than age *per se*, it may be the way age-related changes in health functioning, social losses, and approaching death are dealt with that could be important for the development of spirituality among older adults. Spiritual development is undoubtedly a lengthy maturational process that occurs over the course of adult life when individuals experience the ambiguity and the relativity of human life (Moberg, 2001).

Our results also confirm that older women would have higher scores of spirituality than older men (Wink and Dillon, 2002; Maselko and Kubzansky, 2006; Brown *et al.*, 2013). It is noteworthy that the effect of sex on spirituality scores is strong and clinically meaningful, as indicated by the unstandardized estimate of sex effect and the raw standard deviation of the spirituality measure. Moreover, not only did the older women have higher levels of spirituality than older men, but this upward trend also continued over time. In many studies, gender differences in spirituality have been assumed to exist based on the gender differences rather than sex differences (Bryant, 2007). The main reasons for this difference would be the tendency for women to be more emotionally expressive and they also use more emotion-focused coping styles than men (Goldshmidt and Weller, 2000). Concerning the role of religion in spirituality our result indicates that older adults who had a religion reported higher levels of spirituality than those who had no religion. Based on the unstandardized estimate of religion and the raw standard deviation of spirituality, it can be deduced that the religion effect is strong and meaningful. The relationship between spirituality and religion is not surprising and has already been demonstrated. Although all humans may have a spiritual dimension, religious people could be more predisposed and more aware of this spirituality.

Our second objective was to explore the within-person synergies of spirituality, satisfaction with social support, and flexibility. We hypothesized that trajectories of spirituality were related to satisfaction with social support and flexibility. Our

results indicate that older adults who reported higher levels of social support and flexibility also reported higher levels of spirituality. In addition, the increase in spirituality seems to have accelerated at time points at which participants also had higher rather than lower levels of social support and flexibility. Based on the unstandardized estimates and the raw standard deviation of spirituality, it is noteworthy that these effects are rather small but of theoretical interest. Among dimensions of spirituality, it has been assumed that connection to others and a sense of belonging give meaning to life. In addition, in the framework of aging theory, being in a relationship is an essential dimension of well-being in aging processes (Carstensen *et al.*, 2003; Tornstam, 2005). For example, in the socioemotional selectivity theory (SST, Carstensen *et al.*, 2003), even if there is an age-associated reduction in social networks, older adults prioritize the emotional significance of their relationships to give meaning in life. This explains why when satisfaction in social support increases the sense of life, spirituality also increases. In accordance with Erikson's (1987) theory, old age is defined as the generativity stage. This stage is revealed by a growing concern for well-being of the next generation or society as a whole, expressing a shift away from purely personal interests to more enduring sources of meaning. This suggests that giving support (i.e. an adult's ability to care for another person) may be as important as receiving support. In a longitudinal study, Dillon *et al.* (2003) found a relationship between spirituality and generativity in late adulthood, confirming the link between spirituality and connection to others. A new finding has been revealed in the present study concerning the relationship between flexibility and spirituality. Older adults who reported higher levels of flexibility also reported higher levels of spirituality. Moreover, the linear increase in spirituality seems to have accelerated at time points at which participants also had higher levels of flexibility. An important aspect of adjustment to aging is the ability to realize the positive values of life and its meaningfulness. Flexible adjustment helps to regain an overall sense of efficacy by finding more positive meaning in losses and by reinterpreting the biographical past in self-enhancing ways (Brandtstädter *et al.*, 2010; Bailly *et al.*, 2014). Therefore, to be adaptive in old age, people need to embrace flexibility and to accept change. Our results highlight that spirituality and flexibility undoubtedly play key roles in well-being and adaptive processes to the challenges of aging.

Although this study provides additional information on aging processes, some methodological

limitations should be mentioned. First, although analysis of the potential impact of attrition revealed no major significant differences between participants, some data are missing due to death and differential-losses-to-follow-up that could have biased the results. The second limitation is the degree to which these results can be generalized. Our sample was similar to French national averages (INSEE, 2005) in terms of gender, marital status, and religious affiliations, but respondents had generally completed more years of education than expected for people in this age bracket. In addition, our participants lived at home independently with a good self-health evaluation, few diseases and a high evaluation of financial satisfaction. These characteristics lead us to assume that they did not have severe functional disabilities. A more representative sampling (with more clinical data, such as functional capacity, medication, or cognitive impairment) should be considered to improve understanding of the change in spirituality over time. In addition, our study concerned only a French aging population. France is a secular country and a cross-cultural perspective is necessary. The third limitation concerns the measure of spirituality. Like other instruments used to measure spirituality, DSES might be contaminated with items assessing well-being. As a consequence, it may be more relevant to view DSES as a spiritual well-being measure rather than a spiritual measure. In addition, DSES is a short measure of spirituality. A multidimensional instrument of spirituality (e.g. spiritual attitudes and involvement list) could offer new insights in how social support and flexibility are related with distinct aspects of spirituality. Finally, although DSES can be applied to religious as well as non-religious populations, some items refer to God. Spirituality is a personal and subjective experience and is not easy to measure, and the integration of qualitative approaches will provide complementary understanding of spirituality. The fourth limitation concerns the use of MGCA. Even if our results highlighted relationships between spirituality and social support and flexibility, MGCA do not specify causal relationships among studied variables. It is difficult to say whether individuals are flexible (or satisfied with their social support) because they are spiritual persons or whether they are spiritual persons because they are flexible (or satisfied with their social support). In our view, flexibility and social support would impact on spiritual development, which in turn would impact on flexibility and social support. Finally, given the close relationship between spirituality and well-being quality of life (Koenig *et al.*, 2001; Hill and Pargament, 2008; Ballew *et al.*, 2012; Ali *et al.*,

2015), further investigations need to incorporate well-being outcomes.

In the current demographic aging context, most studies point to a very steep increase in the dependency ratio between 2020 and 2030, implying the need for a better understanding of how people can maintain a positive outlook on their life when resources and control decrease. From this perspective, the results of the present study help to enhance the understanding of the potential benefit of encouraging the spiritual aspects of life, and we hope this will be used in other health-related research.

Conflict of interest

None.

Description of authors' roles

N. Bailly supervised the data collection and drafted the paper. G. Martinent was responsible for the statistical analyses. C. Ferrand and G. Martinent assisted with writing the paper. C. Giraudeau and O. Agli contributed in formulating the research question and to interpreting the results. N. Roussiau and K. Gana had critically revisited the paper and approved it. All authors have read and approved the final version of the paper.

References

- Alaphilippe, D., Bailly, N., Gana, K. and Martin, B.** (2005). Les prédicteurs de l'adaptation chez l'adulte âgé. *L'Année Psychologique*, 105, 649–667. doi:10.3406/psy.2005.30496.
- Ali, J., Marhemat, F., Sara, J. and Hamid, H.** (2015). The relationship between spiritual well-being and quality of life among elderly people. *Holistic Nursing Practice*, 29, 128–135. doi:10.1097/HNP.0000000000000081.
- Bailly, N., Gana, K., Hervé, C., Joulain, M. and Alaphilippe, D.** (2014). Does flexible goal adjustment predict life satisfaction in older adults? A six-year longitudinal study. *Aging & Mental Health*, 18, 662–670. doi:10.1080/13607863.2013.875121.
- Bailly, N. and Roussiau, N.** (2010). The Daily Spiritual Experience Scale (DSES): validation of the short form in an elderly French population. *Canadian Journal on Aging*, 29, 223–231. doi:10.1017/S0714980810000152.
- Ballew, S. H., Hannum, S. M., Gaines, J. M., Marx, K. A. and Parrish, J. M.** (2012). The role of spiritual experiences and activities in the relationship between chronic illness and psychological well-being. *Journal of Religion and Health*, 51, 1386–1396. doi:10.1007/s10943-011-9498-0.
- Bates, D., Maechler, M., Bolker, B. and Walker, S.** (2015). Fitting linear mixed-effects models using lme4.

- Journal of Statistical Software*, 67, 1–48.
doi:10.18637/jss.v067.i01
- Benyamini, Y., Idler, E. L., Leventhal, H. and Leventhal, E. A.** (2000). Positive affect and function as influences on self-assessments of health: expanding our view beyond illness and disability. *The Journals of Gerontology. Series B, Psychological Sciences and Social Sciences*, 55, 107–116.
- Brandtstädter, J. and Renner, G.** (1990). Tenacious goal pursuit and flexible goal adjustment: explication and age-related analysis of assimilative and accommodative strategies of coping. *Psychology and Aging*, 5, 58–67.
doi:10.1037/0882-7974.5.1.58.
- Brandtstädter, J. and Rothermund, K.** (2002). The life-course dynamics of goal pursuit and goal adjustment: a two-process framework. *Developmental Review*, 22, 117–150. doi:10.1006/drev.2001.0539.
- Brandtstädter, J., Rothermund, K., Kranz, D. and Kühn, W.** (2010). Final decenterations: personal goals, rationality perspectives, and the awareness of life's finitude. *European Psychologist*, 15, 152–163.
doi:10.1027/1016-9040/a000019.
- Brown, I. T., Chen, T., Gehlert, N. C. and Piedmont, R. L.** (2013). Age and gender effects on the Assessment of Spirituality and Religious Sentiments (ASPIRES) scale: a cross-sectional analysis. *Psychology of Religion and Spirituality*, 5, 90–98. doi:10.1037/a0030137.
- Bryant, A. N.** (2007). Gender differences in spiritual development during the college years. *Sex Roles*, 56, 835–846. doi:10.1007/s11199-007-9240-2.
- Carstensen, L. L., Fung, H. H. and Charles, S. T.** (2003). Socioemotional selectivity theory and the regulation of emotion in the second half of life. *Motivation and Emotion*, 27, 103–123. doi:10.1023/A:1024569803230.
- Cohen, S., Underwood, L. and Gottlieb, B.** (eds.) (2000). *Social Support Measurement and Interventions: A Guide for Health and Social Scientists*. New York: Oxford.
- Dalby, P.** (2006). Is there a process of spiritual change or development associated with ageing? A critical review of research. *Aging & Mental Health*, 10, 4–12.
doi:10.1080/13607860500307969.
- Dillon, M., Wink, P. and Fay, K.** (2003). Is spirituality detrimental to generativity? *Journal for the Scientific Study of Religion*, 42, 427–442. doi:10.1111/1468-5906.00192.
- Erikson, E. H.** (1987). The human life cycle. In S. Schlein (ed.), *A Way of Looking at Things: Selected Papers from 1930 to 1980 by Erik H. Erikson*. New York: Norton.
- Goldshmidt, O. T. and Weller, L.** (2000). 'Talking emotions': gender differences in a variety of conversational contexts. *Symbolic Interaction*, 23, 117–134. doi:10.1525/si.2000.23.2.117.
- Hill, P. C. and Pargament, K. I.** (2008). Advances in the conceptualization and measurement of religion and spirituality: implications for physical and mental health research. *Psychology of Religion and Spirituality*, 5, 3–17.
doi:10.1037/1941-1022.S.1.3.
- INSEE** (2005). *Les personnes âgées*. Paris, France: INSEE.
- Koenig, H. G., McCullough, M. E. and Larson, D. B.** (2001). *Handbook of Religion and Health*. New York, NY: Oxford University Press.
- Koenig, H. G., Westlund, R. E., George, L. K., Hughes, D. C., Blazer, D. G. and Hybels, C.** (1993). Abbreviating the duke social support index for use in chronically ill elderly individuals. *Psychosomatics*, 34, 61–69. doi:10.1016/S0033-3182(93)71928-3.
- Maselko, J. and Kubzansky, L. D.** (2006). Gender differences in religious practices, spiritual experiences and health: results from the US general social survey. *Social Science & Medicine* (1982), 62, 2848–2860.
doi:10.1016/j.socscimed.2005.11.008.
- Moberg, D. O.** (ed.) (2001). *Aging and Spirituality*. New York: Haworth.
- Seeman, T. E.** (2000). Health promoting effects of friends and family on health outcomes in older adults. *American Journal of Health Promotion*, 14, 362–370.
- Singer, J. D. and Willett, J. B.** (2003). *Applied Longitudinal Data Analysis: Methods for Studying Change and Event Occurrence*. New York: Oxford University Press.
- Tomás, J. M., Sancho, P., Galiana, L. and Oliver, A.** (2016). A double test on the importance of spirituality, the 'forgotten factor', in successful aging. *Social Indicators Research*, 127, 1377–1389. doi:10.1007/s11205-015-1014-6.
- Tornstam, L.** (2005). *Gerotranscendence: A Developmental Theory of Positive Aging*. New York, NY: Springer Publishing Company, Inc.
- Underwood, G. and Teresi, J. A.** (2002). The daily spiritual experience scale: development, theoretical description, reliability, exploratory factor analysis, and preliminary construct validity using health-related data. *Annual Behavioral Medicine*, 24, 22–33.
- Wink, P. and Dillon, M.** (2002). Spiritual development across the adult life course: findings from a longitudinal study. *Journal of Adult Development*, 9, 79–94.
doi:10.1023/A:1013833419122.
- Zimmer, Z., Jagger, C., Chiu, C.-T., Ofstedal, M. B., Rojo, F. and Saito, Y.** (2016). Spirituality, religiosity, aging and health in global perspective: a review. *SSM – Population Health*, 2, 373–381.
doi:10.1016/j.ssmph.2016.04.009.

REVIEW

Strategies employed by older people to manage loneliness: systematic review of qualitative studies and model development

Kalpa Kharicha,¹ Jill Manthorpe,² Steve Iliffe,¹ Nathan Davies¹ and Kate Walters¹

¹Department of Primary Care and Population Health, University College London, Royal Free Campus, Rowland Hill Street, London, NW3 2PF, UK

²Social Care Workforce Research Unit, King's College London, Virginia Woolf Building, 22 Kingsway, London, WC2B 6NR, UK

ABSTRACT

Objectives: To (i) systematically identify and review strategies employed by community dwelling lonely older people to manage their loneliness and (ii) develop a model for managing loneliness.

Methods: A narrative synthesis review of English-language qualitative evidence, following Economic and Social Research Council guidance. Seven electronic databases were searched (1990–January 2017). The narrative synthesis included tabulation, thematic analysis, and conceptual model development. All co-authors assessed eligibility of final papers and reached a consensus on analytic themes.

Results: From 3,043 records, 11 studies were eligible including a total of 502 older people. Strategies employed to manage loneliness can be described by a model with two overarching dimensions, one related to the context of coping (alone or with/in reference to others), the other related to strategy type (prevention/action or acceptance/endurance of loneliness). The dynamic and subjective nature of loneliness is reflected in the variety of coping mechanisms, drawing on individual coping styles and highlighting considerable efforts in managing time, contacting others, and keeping loneliness hidden. Cognitive strategies were used to re-frame negative feelings, to make them more manageable or to shift the focus from the present or themselves. Few unsuccessful strategies were described.

Conclusion: Strategies to manage loneliness vary from prevention/action through to acceptance and endurance. There are distinct preferences to cope alone or involve others; only those in the latter category are likely to engage with services and social activities. Older people who deal with their loneliness privately may find it difficult to articulate an inability to cope.

Key words: ageing, loneliness, qualitative

Introduction

Loneliness is a subjective and distressing experience arising from inadequate social relationships, about which much has been written (see Weiss, 1973; Peplau and Perlman, 1982; Andersson, 1998). It has been characterized in terms of frequency, severity and duration of episode, and illustrating the heterogeneity of the loneliness experience (Victor *et al.*, 2005).

The links between loneliness and its harmful physical and mental health correlates have been

the subject of much research (Luanaigh and Lawlor, 2008; Holt-Lunstad *et al.*, 2010; Valtorta *et al.*, 2016). Efforts to alleviate loneliness have largely focused on interventions to increase contact with others and several systematic reviews have reported on the effectiveness of interventions on loneliness and social isolation. The majority is limited to quantitative outcome studies (Cattan and White, 1998; Findlay, 2003; Cattan *et al.*, 2005; Dickens *et al.*, 2011; Hagan *et al.*, 2014; Cohen-Mansfield and Perach, 2015), and have produced some contradictory and inconclusive findings (Windle *et al.*, 2011). A recent integrative mixed-methods review reiterated the poor quality of the evidence base and called for more qualitative studies to understand the mechanisms underlying successful interventions (Gardiner *et al.*, 2016).

Correspondence should be addressed to: Kalpa Kharicha, Department of Primary Care and Population Health, University College London, Royal Free Campus, Rowland Hill Street, London NW3 2PF, UK. Phone: 020 7830 2392. Email: k.kharicha@ucl.ac.uk Orcid: 0000-0002-2975-2084. Received 1 Dec 2017; revision requested 14 Dec 2017; revised version received 19 Feb 2018; accepted 20 Feb 2018. First published online 25 May 2018.

These efforts are not reflected in the proportion of older people reporting loneliness over the last few decades (Victor *et al.*, 2002; Honigh-de Vlaming *et al.*, 2014); European studies have reported either a small decrease or no change (Dykstra, 2009) and a recent American survey reported an increase in loneliness over the last decade (Wilson and Moulton, 2010).

The subjective nature of loneliness pertains not only to how loneliness is experienced but also to how people respond to and cope with the feelings (Hauge and Kirkeveld, 2013). There is little research into broader strategies older people employ to cope with feelings of loneliness. We have reported the private nature of loneliness and the desire to manage these feelings without involvement of others (Kharicha *et al.*, 2017). This may be a matter of personal preference or due to the stigma of admitting to loneliness (Mental Health Foundation, 2010). Qualitative studies are key to understanding not only the type, but also the extent of support lonely older people might want and would accept. The aim of this review is to (i) systematically review qualitative data to identify strategies employed by community dwelling lonely older people to manage feelings of loneliness themselves, and (ii) develop a model for managing loneliness.

Methods

A systematic review of qualitative studies was carried out using a narrative synthesis approach and followed guidance from the Economic and Social Research Council (ESRC) methods program (Popay *et al.*, 2006), using the stages and tools relevant for this review. The individual stages are outlined in further detail below.

The following databases were searched: Medline, Embase, PsychInfo, CINAHL, WoS, Social policy and practice, ASSIA. Search terms addressing three areas: (i) older people, (ii) social isolation and loneliness, and (iii) coping strategies, were developed iteratively and Mesh terms were used where available (see Appendix 1). The search was run in January 2017 and included papers in the English language from 1990 to January 2017.

Papers were included if participants were aged 65 years and over, were identified or self-identified as lonely, living in the community (including assisted housing arrangements/supported care), reported coping strategies for loneliness, and in which loneliness is the focus of the study. Papers were excluded if study participants were living in residential/nursing care/long-term care facilities or in hospital settings, and those who were terminally ill/receiving palliative care.

Database searches identified 3,043 records and no additional papers were identified from reference lists of included papers or citation tracking. After de-duplicating records, the lead author (KK) reviewed 2,398 titles/abstracts and identified 52 papers for full-text review. A random sample of over 10% of full papers was reviewed by second reviewer (ND). Eligibility of final papers and any papers where there was disagreement were discussed with all co-authors and a consensus reached.

The narrative synthesis approach included the following. A preliminary synthesis extracted relevant data into a pre-defined table and enabled brief textual description of the 11 studies. Data extracted included author, year, country, study design, number/types of participants, analysis, and main themes related to older people's views of coping with loneliness (see Table 1). The Critical Appraisal Skills Programme tool (Critical Appraisal Skills Programme [CASP], 2017) was used to critically reflect on the included studies, but not as a basis to exclude studies. Thematic analysis of text (Strauss and Corbin, 2008) from the findings and conclusions of the papers was carried out; authors' comments in discussion sections were not included in data extraction or synthesis. Finally, a conceptual model was developed by all co-authors to visually represent the relationship between key emergent themes from the review (Popay *et al.*, 2006). This review paper addresses the 21 items in the guidelines for enhancing transparency in reporting the synthesis of qualitative research (ENTREQ) (Tong *et al.*, 2012).

Results

Description of studies included

A total of 11 eligible papers were identified as shown in the PRISMA flow chart (see Figure 1), and a summary of each paper is reported in Table 1.

Data from a total of 502 participants are reported predominantly from interviews or from focus groups. Sample sizes varied from 12 to 170 reflecting the data collection method used. Nine papers reported primary analysis of data and two reported secondary analysis (Kirkeveld *et al.*, 2013; Sullivan *et al.*, 2016) with one of these combining both secondary and primary analyses of data (Sullivan *et al.*, 2016). Two studies also collected professionals' views (Cattan *et al.*, 2003; Stanley *et al.*, 2010); these findings were reported separately in the papers and were excluded from this review. The nine papers reporting primary research all used one-to-one, face-to-face interviews, either semi-structured or in-depth. In addition, two also used focus groups (Cattan *et al.*, 2003; Stanley *et al.*,

Table 1. Description of studies included, in chronological order, *n* = 11

| AUTHORS | YEAR/COUNTRY | STUDY DESIGN | NUMBER / TYPE OF PARTICIPANTS | ANALYSIS | MAIN THEMES RELATED TO OLDER PEOPLE'S VIEWS OF COPING WITH LONELINESS |
|--|----------------|---|--|---------------------------------------|--|
| 1 Cattan, M., Newell, C., Bond, J. and White, M. | 2003/England | Semi-structured interviews & focus groups | 23 staff members from voluntary sector projects targeting loneliness and social isolation in older people, 22 focus groups with 145 older people who participated in project activities, 25 interviews with older people whom project staff considered to be socially isolated and lonely. Older people were 55–94 years old | Framework analysis | (i) Perceptions and experiences of social isolation and loneliness (ii) Coping strategies (iii) Perceptions and experiences of services and activities (iv) Solutions |
| 2 Pettigrew, S. and Roberts, M. | 2008/Australia | Semi-structured interviews | 19 older people living in retirement villages or on their own aged between 65–95 years | Thematic analysis (though not stated) | (i) Social interaction: (a) Interacting with others, (b) eating and drinking rituals; (ii) solitary activities: (a) reading, (b) gardening, and (c) television. |
| 3 Granheim, U.H. and Lundman, B. | 2010/Sweden | Interviews | 30 people aged 85–103 years old, who lived alone in their own homes or apartments in houses for older people | Content analysis | Themes related to loneliness are intertwined with themes related to experiences of ageing whilst living alone. Four main themes: (i) Living with losses: Suffering from bodily decline; being dependent; mourning significant others; missing zest for life; longing for meaning (ii) Feeling abandoned: Feeling set aside and feeling invisible (iii) Living in confidence: Feeling safe and secure; leaving everything in God's hands; feeling content; and accepting the loneliness (iv) Feeling free: Being able to decide about one's own business; feeling spared from duties and worries; resting in peace and quiet; and having the opportunity to make new friends |

Table 1. Continued

| AUTHORS | YEAR/COUNTRY | STUDY DESIGN | NUMBER / TYPE OF PARTICIPANTS | ANALYSIS | MAIN THEMES RELATED TO OLDER PEOPLE'S VIEWS OF COPING WITH LONELINESS |
|---|-----------------------|---|---|---|--|
| 4 Roos, V. and Klopper, H. | 2010/South Africa | In-depth interviews | 31 older people, 4 lived in residential care, 3 with their children, 1 in own home, the rest ($n = 23$) in own house/flat in a retirement village. 16 Afrikaans speaking had mean age 79 years (SD 9.6), 15 English speaking Tswana people had mean age 70 years (SD 6.0) | Phenomenological approach to identifying themes | 3 themes: expressions of loneliness, causes of loneliness, and coping with loneliness Coping with loneliness: Self-awareness and preferred style of interaction; humor; preparations for and dealing with losses; meaningful interpersonal contact; religion; and active engagement with life |
| 5 Stanley, M., Moyle, W., Ballantyne, A., Jaworski, K., Corlis, M., Oxlade, D., Stoll, A. and Young, B. | 2010/Australia | Focus groups and semi-structured interviews | 8 focus groups with approximately 64 support/service providers for older people, and interviews with 60 older people living in long-term care, independent living units, and the community, aged between 67 and 92 years | Thematic analysis | Key themes describing loneliness, as: (i) Private, (ii) relational, (iii) connectedness, and (iv) temporal The themes focus on experiences of loneliness with limited accounts of coping strategies |
| 6 Lou, V.W.Q. and Ng, J.W. | 2012/Hong Kong, China | Semi-structured interviews | 13 community dwelling adults, living alone and not severely lonely (on de Jong-Gierveld loneliness scale), aged 62–88 years | Interpretive approach | A cultural specific model: relationship-oriented resilience to senses of loneliness in a Chinese context 3 primary themes: (i) Cognitive resilience: (a) Cognitive pragmatics, (b) everyday competence (ii) Self and personality: (a) Interdependent self, (b) open and accommodating (iii) Social relations: (a) Social affiliation, (b) social companionship |

Table 1. Continued

| AUTHORS | YEAR/COUNTRY | STUDY DESIGN | NUMBER / TYPE OF PARTICIPANTS | ANALYSIS | MAIN THEMES RELATED TO OLDER PEOPLE'S VIEWS OF COPING WITH LONELINESS |
|---|------------------------------|--|---|--|--|
| 7 Smith, J.M. | 2012/USA | Mixed methods (quantitative & semi-structured interview) | 12 people aged between 74 and 98 years old, either attending senior centers or (the majority) via personal contact from colleagues and friends | Interpretative phenomenological approach including thematic analysis | (i) Sustaining connections with others: (a) Reaching out to others, (b) helping those in need (volunteering), (c) seeking companionship with pets (ii) Finding comfort in television and other hobbies |
| 8 Kirkevoid, M., Moyle, W., Wilkinson, C. Meyer, J. and Hauge, S. | 2013/ Australia, Norway & UK | Secondary analysis of in-depth interviews | 78 older people: 15 from Australia (mean age 79 years), 33 from the UK (mean age 81 years) and 30 from Norway (mean age 85 years). A total of 26 lived in long-term care, 19 in an independent living unit, and 33 in private homes | Thematic analysis (though not stated) | Impact of losses was closely related to loneliness experience. Four central themes related to losses were dichotomized by "not lonely"/"lonely" groups as follows: (i) Accepting losses and moving on versus being overpowered by accumulating losses (ii) Staying committed to activities versus unable to carry on with activities (iii) Staying connected to other people versus being isolated from other people (iv) Creating a meaningful life in one's own company versus a life alone is an empty life Three broad themes: (i) Experiencing the absence, (ii) loss of routine connection, and (iii) establishing new routines, which describe the experience of loneliness following widowhood from an acute phase of experiencing an absence and the associated loss of routine connection to the establishment of new routines that provided new connections and a new sense of identity as an individual rather than a couple. The process was facilitated by keeping active and being mobile. The participants also described having to manage the social norms associated with what behaviors others expected from a widow or widower. This was not a linear trajectory of recovery from loneliness and many of the participants continued to experience periods of loneliness |
| 9 Davies, N., Crowe, M. and Whitehead, L. | 2016 / New Zealand | Narrative inquiry | 40 older widow/widowers, aged 70–97 years | Thematic analysis | Three broad themes: (i) Experiencing the absence, (ii) loss of routine connection, and (iii) establishing new routines, which describe the experience of loneliness following widowhood from an acute phase of experiencing an absence and the associated loss of routine connection to the establishment of new routines that provided new connections and a new sense of identity as an individual rather than a couple. The process was facilitated by keeping active and being mobile. The participants also described having to manage the social norms associated with what behaviors others expected from a widow or widower. This was not a linear trajectory of recovery from loneliness and many of the participants continued to experience periods of loneliness |

Table 1. Continued

| AUTHORS | YEAR/COUNTRY | STUDY DESIGN | NUMBER / TYPE OF PARTICIPANTS | ANALYSIS | MAIN THEMES RELATED TO OLDER PEOPLE'S VIEWS OF COPING WITH LONELINESS |
|---|---------------------|---|---|-------------------|--|
| 10 Taube, E., Jakobsson, U., Midlöv, P. and Kristensson, J. | 2016/Sweden | Semi-structured interviews | 12 older people purposively selected from a larger intervention study (randomized controlled trial), aged 68–88 years (mean 79 years). | Content analysis | Overall theme: Being in a Bubble three themes with subthemes within this: (i) Barriers: (a) The ageing body, (b) fear, (c) the influence of losses, and (d) no one to share daily chores with (ii) Hopelessness: (a) A constant state, (b) feeling sad, empty and anxious, (c) being invisible to others, and (d) losing the spirit (iii) Freedom: (a) Having time to reflect and reload, (b) being free to make decisions, (c) being able to create meaningfulness, (d) having a social belonging, and (e) being protected from disappointment Findings separate the accounts of those who talked “openly” about loneliness from those who found it harder to talk about, emphasizing the dynamic and multi-dimensional aspects of loneliness Coping strategies include internal factors e.g. acceptance, finding, inner strength, keeping loneliness hidden, and external factors e.g. reading, “keeping busy,” having routines, maintaining and activating social networks |
| 11 Sullivan, M.P., Victor, C.R. and Thomas, M. | 2016/England, UK | Secondary analysis of in-depth interviews from study 1 informed the development of primary data collection using in-depth interviews in Study 2 | Study 1. 25 people aged 67–87 years (mean 81 years) who were lonely/sometimes lonely in a larger mixed methods study on loneliness and social isolation in later life Study 2. 12 people aged 65–81 years (mean 71 years) participating in a mixed methods pilot longitudinal study on temporal variations in loneliness | Thematic analysis | |

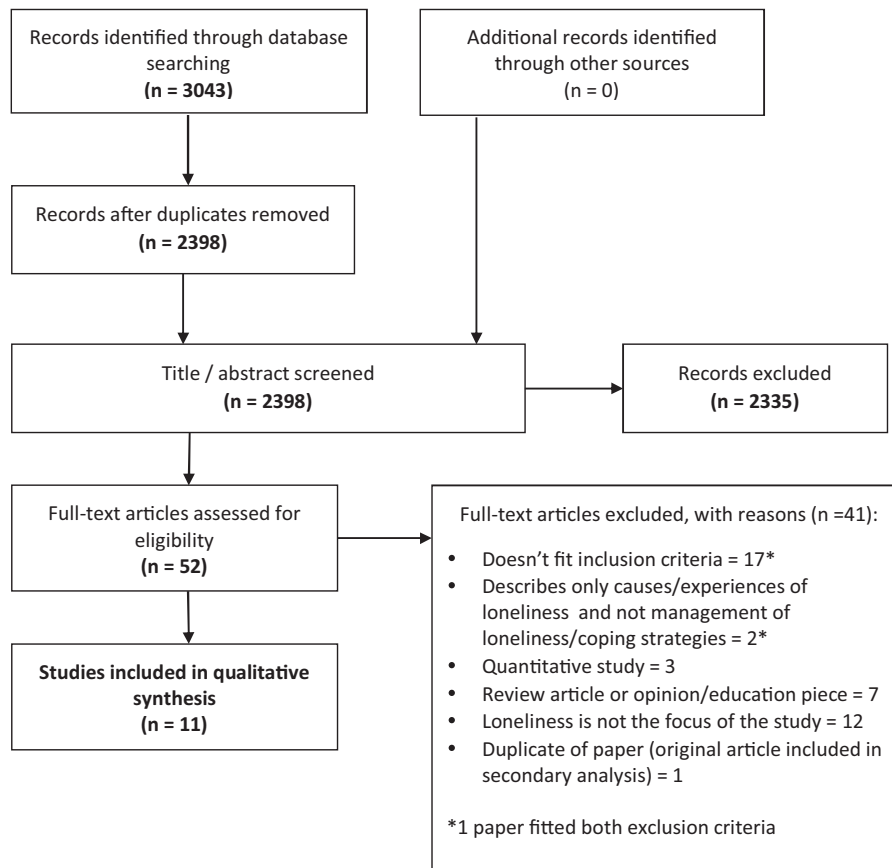


Figure 1. PRISMA flow chart.

2010), while another was a mixed method study collecting additional quantitative data (Smith, 2012).

All studies included participants aged 65 year and over, although age ranges varied from 55–94 years (Cattan *et al.*, 2003) to 85–103 years (Graneheim and Lundman, 2010). Similarly, all studies included community dwelling older people living either in their own homes or living independently in retirement villages or independent living units. Two studies also recruited older people living in more supported housing, including long-term care (Stanley *et al.*, 2010) and residential care facilities (Roos and Klopper, 2010). Findings are not differentiated by age band, the type of housing, or support/care the participants received, including whether participants required assistance to leave their homes.

Quality appraisal

Overall the studies were of mixed quality. Several papers reported both *experiences* of loneliness as well as *responses* to loneliness (e.g. Smith, 2012; Taube *et al.*, 2016; Davies *et al.*, 2016; Sullivan *et al.*, 2016) and in two papers the data on responses were

particularly limited (Cattan *et al.*, 2003; Stanley *et al.*, 2010). Two papers used the term social isolation interchangeably with loneliness (Cattan *et al.*, 2003; Pettigrew and Roberts, 2008). Other papers categorized participants as being “lonely” or “not lonely” and inferred that strategies described by those who were “not lonely” were potential strategies to prevent loneliness (e.g. Lou and Ng, 2012; Kirkevold *et al.*, 2013). The implications of how lonely older people are identified were important and are discussed below. These papers were included in this review, but the contribution of the data is limited accordingly.

Recruitment in primary studies was reported in varying detail. Some employed multiple strategies reflecting the potential difficulty in recruiting lonely older people to loneliness research. Information on notice boards (Roos and Klopper, 2010) or newsletters and flyers (Stanley *et al.*, 2010) was used alongside asking key contacts within organizations to recruit potential participants. Recruitment via professionals was common (Cattan *et al.*, 2003; Pettigrew and Roberts, 2008; Roos and Klopper, 2010; Stanley *et al.*, 2010; Lou and Ng, 2012; Smith, 2012; Davies *et al.*, 2016). One study changed its recruitment approach after failing to

recruit sufficiently (Smith, 2012). Only Taube *et al.* (2016), who recruited from a larger intervention study, report any detail of response rates. The papers reporting secondary analyses refer to the original sources of data and little can be gleaned about recruitment from reading the two papers alone.

There was no direct reference to reflexivity although two studies (Davies *et al.*, 2016; Taube *et al.*, 2016) described researchers' professional backgrounds and any previous knowledge of the study participants. One (Roos and Klopper, 2010) recognized the importance of being wary of researchers' views of loneliness while interviewing. Two studies (Cattan *et al.*, 2003; Pettigrew and Roberts, 2008) involved participants in respondent validity (by sharing transcripts or early themes) but did not report if the analysis or interpretation were shaped by this. Two papers referred to having used the COREQ checklist (Tong *et al.*, 2007) for reporting (Davies *et al.*, 2016; Taube *et al.*, 2016).

Results were well mostly well-presented with core themes and verbatim quotes. However, authors' reflections were not consistently backed by data (Sullivan *et al.*, 2016), or quotes were merged within the descriptive text without accompanying demographic data for information or to gauge the spread of participants' views (Roos and Klopper, 2010). Two papers reported the hierarchy of themes (Roos and Klopper, 2010; Taube *et al.*, 2016). However, one paper combined data from older people attending community groups, and those attending who were thought to be lonely by the staff, not differentiating between the two data sets in their findings (Cattan *et al.*, 2003).

Culturally bound interpretations, specific to Hong Kong Chinese and South African culture, are reported by Lou and Ng (2012) and Roos and Klopper (2010), respectively, but there is no discussion of cultural differences in the secondary analysis of the three-country dataset by Kirekevold *et al.* (2013).

Identifying older people who are lonely

Four different approaches were used to identify older people who might be lonely, summarized in bold text here (most studies used more than one approach). First, older people **self-identified** as lonely in 3 of the 11 studies. Smith (2012) interviewed those responding positively to the question: "Have you experienced loneliness within the last six months?" although it is unclear whether this initial question was asked verbally or presented in written form. Others had reported being "lonely," "sometimes lonely" or given an indication of strength of loneliness feelings in an earlier study from which they were then purposively

sampled for interview (Taube *et al.*, 2016; Sullivan *et al.*, 2016) or reported being lonely or sometimes lonely during an interview (Sullivan *et al.*, 2016).

In four studies, participants had not necessarily identified themselves as lonely but simply reported that they were **willing to talk about loneliness**, (Pettigrew and Roberts, 2008; Roos and Klopper, 2010; Stanley *et al.*, 2010; Kirekevold *et al.*, 2013). Several studies (6 out of 11) **relied on practitioners** at community organizations running groups or activities for older people, general practices, or elder care/retirement village managers, to identify potential participants, that is, older people they thought were lonely or at risk of loneliness and likely to be interested in participation (Cattan *et al.*, 2003; Pettigrew and Roberts, 2008; Roos and Klopper, 2010; Lou and Ng, 2012; Smith, 2012; Davies *et al.*, 2016). Lou and Ng's (2012) culturally specific approach to coping strategies for loneliness was the only study to use a validated loneliness measure (de Jong-Gierveld scale) (de Jong-Gierveld, 1987). They excluded those whose scores indicated severe loneliness and included all others who were hence considered to be coping with loneliness as they did not report being severely lonely despite living alone. Finally, in 8 of the 11 papers, a range of "**risk factors**" was **used as proxy measures** to identify loneliness. These included being widowed (Davies *et al.*, 2016) or being very old (85 years and over) and living alone (Graneheim and Lundman, 2010), attending community groups/day centers or those living in retirement villages (Cattan *et al.*, 2003; Pettigrew and Roberts, 2008; Roos and Klopper, 2010; Stanley *et al.*, 2010; Lou and Ng, 2012; Smith, 2012; Davies *et al.*, 2016). These participants may or may not have been lonely themselves; they often talked of "others" rather than themselves. Furthermore, Sullivan *et al.* (2016) reported that some who had previously rated themselves as lonely did not go on to volunteer this when interviewed.

Findings of the synthesis

The thematic analysis identified strategies employed by older people to manage their loneliness. The themes can be grouped into two overarching themes or dimensions. The first dimension relates to context and describes whether people cope (and choose to manage) alone or prefer to cope with/in reference to others (that is, with others in mind). The second dimension refers to the type of strategy employed, and represents a spectrum ranging from prevention or action in response to loneliness or acceptance or endurance of loneliness. Prevention of loneliness describes both the strategies participants reported they would put

in place if they felt lonely, as well as actual strategies practised in an attempt to deter loneliness. The former “hypothetical actions” may arise as a result of how participants were identified (as discussed above) and the uncertainty in whether or not they were in fact lonely, and/or the difficulties of disclosing or describing feelings of loneliness. Actions were the strategies people described they performed to alleviate their distress. Acceptance and endurance of loneliness overlap to some extent in their definition but differ in that “acceptance” is taken to mean an adequate resolution to the experience of loneliness and “endurance” that the unpleasant feelings continue and are “lived with.” The two dimensions can be represented as a model of managing loneliness, as presented in the *Discussion* section of this paper (see Box 1).

The findings of the synthesis are presented below, with themes grouped within the two overarching dimensions, as appropriate. Some themes, including personality-related factors, the effort involved in planning, cognitive strategies, and going outdoors, are mentioned more than once as they describe strategies that can be placed within both dimensions. Verbatim participant quotes are used to illustrate themes where possible from papers that reported primary data.

Coping alone

A range of factors were identified across studies that supported coping alone with loneliness.

Prevention and action

- *Personality-related strategies* included being determined and motivated to stay active, focusing on good times, taking pride in yourself and your environment, the ability to shift the focus away from yourself, and onto the outside world and finding humor in situations (Roos and Klopper, 2010; Stanley *et al.*, 2010; Lou and Ng, 2012; Kirkevold *et al.*, 2013), as this quotation illustrated.
- “You have to keep pushing yourself all the time. I am actually conscious of not sitting in my chair. I have to keep getting up and doing something” (Stanley *et al.*, 2010, p. 410).
- The driver for these strategies was the belief that it is an individual's responsibility to manage their feelings of loneliness (Roos and Klopper, 2010; Stanley *et al.*, 2010; Sullivan *et al.*, 2016) and a lack of response would mean they could possibly lead to depression or worsen it (Roos and Klopper, 2010). Some personality-related strategies may have been lifelong traits while others were age related, for example, feeling a “freedom of expression” in later life that allows a license to behave differently (Taube *et al.*, 2016).

- The efforts involved in establishing and maintaining plans, structure, and routines were described by many, often in an effort to fill the time. This was in relation to daily structures as well as re-establishing routines and adjusting after significant life events and losses and planning for inevitable loneliness (Roos and Klopper, 2010; Lou and Ng, 2012; Kirkevold *et al.*, 2013; Sullivan *et al.*, 2016; Davies *et al.*, 2016). A common element within daily routines was *going outdoors regularly* for stimulation (e.g. Roos and Klopper, 2010; Lou and Ng, 2012).
- The idea of “*keeping busy*” was mentioned in most accounts. Solitary pastimes ranged from activities, interests, and hobbies that were considered more engaging or “meaningful” than others such as reading, gardening, walking, and following current events, compared with those that were considered a distraction or more “passive” such as watching TV other than the news (Pettigrew and Roberts, 2008; Roos and Klopper, 2010; Lou and Ng, 2012; Smith, 2012; Kirkevold *et al.*, 2013; Taube *et al.*, 2016; Sullivan *et al.*, 2016). As one study participant reported: “... I keep busy and I don't get lonely” (Kirkevold *et al.*, 2013 p. 397).
- *Religion, spirituality, and philosophical approaches* were raised in papers reporting the experiences of the very old living alone and of a South African older population. Having a religion or faith and a belief that you are not alone as God is with you, in life as well as death, made them less fearful. Spiritual practices reported included prayer, singing, and reading alone as well as engaging in meditation or “forced calmness” (Graneheim and Lundman, 2010; Roos and Klopper, 2010), as illustrated by one study participant: “I know I'm never alone, the Lord is always with me” (Roos and Klopper, 2010, p. 286).

Acceptance and endurance

1. *Loneliness as inevitable.* Perceiving loneliness as inevitable, commonplace, and experienced by all was a way of coming to terms with feelings of loneliness and accepting them (Pettigrew and Roberts, 2008; Graneheim and Lundman, 2010; Roos and Klopper, 2010).
2. *Personality-related strategies* relating to an acceptance or endurance of loneliness portrayed a positive attitude, an ability to draw on “inner strength,” a sense of control over one's experience of loneliness and the extent to which it is experienced (Roos and Klopper, 2010; Sullivan *et al.*, 2016). For the very old this was described as having a “fateful” approach and living in the moment, being happy for each new day and not wanting more: “Yes, you should take everything as it comes ... nothing is that important ... I am just a little dot in the universe and still I am wonderful ... a wonderful creation” (Graneheim and Lundman, 2010, p. 436).

3. Acknowledging the *temporal nature of loneliness* helped people cope. Learning from previous episodes of loneliness and understanding that both the episode and how acutely it is felt can pass (Roos and Klopper, 2010; Stanley *et al.*, 2010; Sullivan *et al.*, 2016). The strategies were not described as “cures” for loneliness, and there was no sense of permanent resolution. Rather they were strategies that could be drawn on to bring temporary relief to feelings of loneliness which came and went at different times of day, week or season, after specific life events and over their life course and varied in intensity at different times. One paper summarized such a situation thus: “... he felt lonely at night after his wife had gone to bed, but his salvation was his reading – this time provided an opportunity for him to become aware of his loneliness but able to temporarily escape it ...” (Sullivan *et al.*, 2016, p. 174).
4. Another strategy was *comparative thinking* in which people found some relief by comparing their current situation and feelings to times of life that had been more difficult emotionally, for example, when younger (Graneheim and Lundman, 2010; Lou and Ng, 2012; Sullivan *et al.*, 2016).
5. *Re-framing loneliness* to consider the advantages of being alone was reported by some who focused on their time alone as an opportunity to reflect and rest, or enjoy the freedom to do what one wanted and a pride in one’s ability to live alone in later life (Graneheim and Lundman, 2010; Roos and Klopper, 2010; Taube *et al.*, 2016), as mentioned by this one study participant: “When you’re alone and have chosen to be alone. When you think, ‘oh, how nice it is to sit here’” (Taube *et al.*, 2016, p. 637).
6. For loneliness that is private and persistent, one paper used the metaphor of “fighting” to describe the *constant effort to fight the feelings* of loneliness, including an acknowledgment of its persistence, and efforts to find small relief where possible (Taube *et al.*, 2016).

Coping with/in reference to others

In this section, we describe strategies identified from studies on coping through the involvement of other people.

Prevention and action

1. *Establishing, maintaining, nurturing, repairing relationships, and connections* throughout life were described in most papers. This most commonly referred to family and friends, but also pets (Smith, 2012) and care workers (Graneheim and Lundman, 2010). These contacts took place both inside and outside the home (including the use of the telephone), and were for social and/or emotional loneliness, that is from “simple” contact to having confidantes. As one paper noted, this could be routinized: “I look forward to being

able to wander over there (the retirement village’s communal lounge area) at 5 o’clock each night and be able to sit and have a couple of drinks for an hour and then come home and have tea” (Pettigrew and Roberts, 2008, p. 306).

Within this was a sense of hierarchy of relationship between families and friends, as well as the need for both, while having boundaries around what is shared within these groups in order to maintain the relationship(s) (Pettigrew and Roberts, 2008; Roos and Klopper, 2010; Stanley *et al.*, 2010; Lou and Ng, 2012; Smith 2012; Kirkevold *et al.*, 2013; Davies *et al.*, 2016; Sullivan *et al.*, 2016; Taube *et al.*, 2016).

2. The *effort to plan and initiate arrangements* in reference to “others” most commonly occurred over food and drink rituals, both more “formal” meals or special occasions that may have been practised throughout life and “informal” exchanges such as “having a drink” (alcohol), “going for coffee,” “having tea,” which were often culturally bound (Pettigrew and Roberts, 2008; Lou and Ng, 2012; Smith, 2012). People also described having plans or “back up” strategies if they were to start feeling lonely (Sullivan *et al.*, 2016) such as this study participant’s practice: “I mean I could always go up and talk to the girl, the sisters, or go and talk to Sam and Catherine. There are several people, if I felt lonely, which I don’t. Or they’d come and see us [...] I mean if I really felt lonely I’d take the dogs over to the common and I’d find someone to talk to very, very quickly” (Sullivan *et al.*, 2016, p. 173).
3. Again, *going outdoors regularly* as a strategy was discussed this time with the hope to initiate or increase the likelihood of chance encounters and exchanges with others (Cattan *et al.*, 2003; Lou and Ng, 2012; Sullivan *et al.*, 2016). One person described this approach: “I try not to get lonely but I do. I go out to try to stop being lonely. I sit and talk to people in the park. I get lonely a lot – that’s why I go out a lot” (Cattan *et al.*, 2003, p. 25).
4. *Shifting the focus away from yourself and onto others* either by engaging in activities that were considered meaningful and worthwhile such as volunteering and caring responsibilities as well as socializing for the sake of others and not just yourself also emerged (Pettigrew and Roberts, 2008; Roos and Klopper, 2010; Smith, 2012; Kirkevold *et al.*, 2013; Taube *et al.*, 2016).
5. *Being open to new experiences* such as clubs or activities to establish contact with others was raised in a few papers and conveyed a reluctant necessity in the description. It required courage and was often challenging. This was sometimes due to a loss of confidence that had developed over time in initiating such contacts or following life events such as bereavement. Those that had tried this approach described it as a “life-line” when there were no other alternatives, where they had found some enjoyment in a safe environment (Cattan

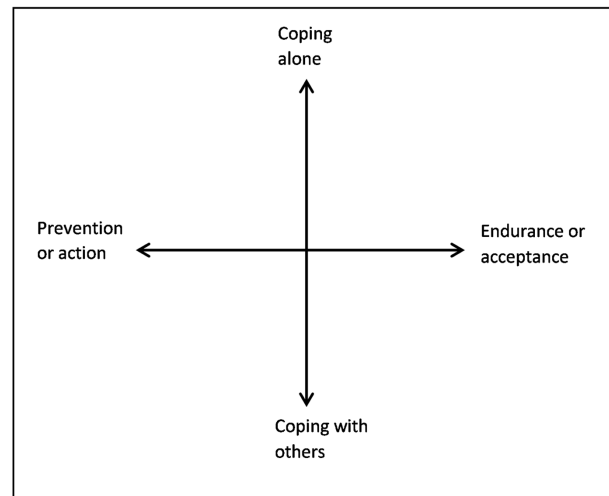
et al., 2003; Lou and Ng, 2012; Kirkevold *et al.*, 2013; Davies *et al.*, 2016), for example, "I go to a club now you see, it's nothing fancy but it serves a purpose ... It took me three or four visits before I started to settle in. We play games and that sort of thing, it takes you mind off things" (Davies *et al.*, 2016, p. 536).

6. Having a *religion or faith and engaging in religious practices* including contact with religious leaders as well as social gatherings helped some and provided particular support after a traumatic event or loss. Religious leaders helped "to pull the wagon through the ditch" (Roos and Klopper, 2010).

Acceptance and endurance

1. *Keeping loneliness hidden or a secret* was described in a few papers. People reported distancing themselves from others or denying their own loneliness and describing the loneliness of "others." This was due to the perception of admission of loneliness as failure and not wanting this to impact on relationships or the difficulties of speaking about loneliness (Stanley *et al.*, 2010; Lou and Ng, 2012; Sullivan *et al.*, 2016). Such a view was expressed by one study participant: "Society sees it as a nasty problem that they don't want to know about and also people who are lonely ... [feel unable] to express this without feeling that they are a failure of some kind" (Stanley *et al.*, 2010, p. 410).
2. *Comparisons* were made either to other people or situations perceived as being "worse" or times when they themselves were "worse off." For example, loneliness may be safer than disappointment, preferring living alone rather than finding a new partner (Cattan *et al.*, 2003; Taube *et al.*, 2016). One participant expressed such wariness thus: "I guess maybe I could have found myself a woman... but... I haven't felt that lonely... I've preferred being alone... If you had a wife who was sick for ten years and it was only trouble, then... for the most part ... you think of that... you don't want to experience that again" (Taube *et al.*, 2016, p. 637).
3. *Taking the focus away from yourself and onto "collective well-being,"* for example, by living alone rather than with families, thus reducing the potential pressures on wider family, was described in a cultural context by Lou and Ng (2012) and without any reference to culture by Kirkevold *et al.* (2013). These papers described an adjustment of expectations to fit with the needs of the wider family, rather than their individual needs.

As might be expected, most of the strategies identified in this review were positively framed. However, a small number of instances were described or alluded to in which people described coping less well, reflecting the fact that it is probably easier to talk about how you cope or would cope, rather than how you might not be managing. These include feelings of desperation



Box 1. A model for managing loneliness in later life.

(Cattan *et al.*, 2003), boredom, gloominess, and feeling abandoned (Graneheim and Lundman, 2010), boredom and meaninglessness (Kirkevold *et al.*, 2013), feeling fearful, vulnerable and hopeless (Taube *et al.*, 2016), descriptions of guilt and shame of not coping, and crying (Roos and Klopper, 2010), heavy alcohol consumption (Pettigrew and Roberts, 2008), and talking about "others" rather than themselves (Sullivan *et al.*, 2016). These have not been included in this analysis as they were only briefly mentioned in the papers.

Discussion

Summary of findings

This review of strategies employed by lonely older people to manage their loneliness identified two main dimensions. First, the context of coping which was either alone or with/in reference to others. Second, the approach to coping which ranged from prevention or action as a response, or acceptance, or endurance of loneliness. The two dimensions can be represented as a model of managing loneliness in later life (see Box 1).

An individual at a given point in time could be placed on these continuums according to their desire to involve others in their loneliness and their preference of strategy type within the spectrum. This model is novel in that it moves beyond understanding the phenomenon of loneliness itself to consider the range of ways older people with loneliness wish to address the issue.

Within the main dimensions, a range of strategies were described, drawing on individual personality-related coping styles, the considerable efforts in managing time, making contact with others, and keeping loneliness hidden. Cognitive

strategies were used to re-frame negative feelings, to make them more manageable or help shift the focus from the present time or themselves. As such these themes may appear within more than one dimension of the model. Difficulty talking about loneliness may account for strategies that might be perceived as being less successful and were infrequently described.

Strengths and limitations of this review

While much has been written about the experience of loneliness in later life and how it might be alleviated, the papers identified in this review of strategies to self-manage loneliness were limited to only 11 in number, all except one having been published within the last 10 years. Over time there appears to have been a slight shift from exploring how services and interventions can help alleviate loneliness to understanding how people manage their distress themselves.

Limitations

Although a systematic approach was taken to this review, it is possible some papers were missed. This review was mainly secondary analysis of primary research based on both verbatim quotes and original author interpretations, but also included papers reporting secondary analysis. The analysis and interpretations reported in this review build on previous interpretations from the authors of the included studies and increasing distance from the views of older people themselves.

Caution is also needed regarding the positive framing of coping with loneliness; it appears as if most lonely people have successful coping strategies. It was not possible to differentiate between those with temporary and chronic loneliness; coping strategies are likely to differ according to length and intensity of loneliness experience. It may also be harder to talk about not coping and those who were struggling may not have volunteered to be interviewed.

Methodological limitations

The range of settings from which older people were recruited meant this review included participants from a wide age range, and varying health needs, living circumstances, and cultural backgrounds. Papers also included both those who self-identified as lonely and those willing to talk about it who may or may not have been lonely. This latter group was often recruited on the basis of a risk factor for loneliness, such as living alone. There were no differentiation or sub-group analyses, although these characteristics may well impact on experience or views of loneliness and/or coping strategies.

Strategies employed by those who were not lonely despite being considered “at risk” were inferred as being protective factors against loneliness. There is an implication that these protective factors could be used as coping strategies by those who were lonely, especially if symptoms of loneliness were “caught early,” including by professionals in contact with these older people. However, this may be more attributable to personality factors, lifelong traits, ways of doing things, and individual coping styles.

Comparison with other literature

The heterogeneity of coping mechanisms for loneliness in later life identified in this review mirrors the breadth of experiences of loneliness that have been reported (Peplau and Perlman, 1982; Andersson, 1998; Victor *et al.*, 2005). Defining loneliness as a discrepancy between actual and desired levels of social engagement (Peplau and Perlman 1982) aligns with a deficit model of ageing. This review identified responses to loneliness, both private and with others, which are largely initiated by an older person suggesting that older people should be regarded as active agents in managing their loneliness (Elder and Johnson, 2003).

Most people experience loneliness at some point in their lives; identifying who might benefit from more intensive psychological support and what this should include is less clear. Services for loneliness currently focus mainly on promoting engagement in group social activities or one-to-one befriending; however, the evidence for effectiveness of existing interventions is mixed (Cattan *et al.*, 2005; Hagan *et al.*, 2014; Cohen-Mansfield and Perach, 2015; Gardiner *et al.*, 2016). There are no interventions to our knowledge currently widely used in practice that explicitly aim to facilitate older people to develop cognitive strategies including acceptance.

Additionally, it has been argued that loneliness is not only about how people view themselves, but also about how they feel they are positioned in society. Societal and community level responses to loneliness include both a normalization of loneliness and manufactured opportunities to re/engage with local communities in later life (Barke, 2017).

Implications

Based on the findings of this review, we have proposed a tentative model to describe ways in which older people may wish to manage their loneliness. Given the dynamic nature of feelings of loneliness it should not be regarded as static. The model has potential for use by practitioners to engage in discussion with lonely older people to

identify ways in which they may want to address their feelings of loneliness drawing on individual coping styles and preferences. Further research is required to explore the acceptability of the model to lonely older people and its applicability to different contexts, settings, and groups.

Conclusions

Based on the findings of this review of qualitative studies, we have developed a model for managing loneliness in later life. The model conceptualizes coping styles for loneliness as being on two-key dimensions representing a spectrum of strategies from prevention or action through to acceptance or endurance, and coping alone or coping with/in reference to others. Older people who choose to deal with their loneliness by themselves may find it difficult to articulate an inability to cope. This taxonomy of coping with loneliness could have implications for interventions to reduce loneliness, if validated by other studies.

Conflicts of interest

None.

Description of author roles

KK designed the review, carried it out, analyzed the data, developed the conceptual model, and led the write up of the paper. JM supervised the review, discussed and agreed eligibility of final papers, contributed to the analysis, conceptual model development, and write up of the paper. SI supervised the review, discussed and agreed eligibility of final papers, contributed to the analysis, conceptual model development, and write up of the paper. ND was second reviewer and reviewed a random sample of papers, discussed and agreed eligibility of final papers, contributed to the write up of the paper. KW supervised the review, discussed, and agreed eligibility of final papers, contributed to the analysis, conceptual model development, and write up of the paper.

References

- Andersson, L.** (1998). Loneliness research and interventions: a review of the literature. *Aging and Mental Health*, 2, 264–272.
- Barke, J.** (2017). Community-based research and approaches to loneliness prevention. *Working with Older People*, 21, 115–123.
- Cattan, M., Newell, C., Bond, J. and White, M.** (2003). Alleviating social isolation and loneliness among older people. *International Journal of Mental Health Promotion*, 5, 20–30. doi:10.1080/14623730.2003.9721909.
- Cattan, M. and White, M.** (1998). Developing evidence-based health promotion for older people: a systematic review and survey of health promotion interventions targeting social isolation and loneliness among older people. *Internet Journal of Health Promotion*, 13, 1–9.
- Cattan, M., White, M., Bond, J. and Learmouth, A.** (2005). Preventing social isolation and loneliness among older people: a systematic review of health promotion interventions. *Ageing & Society*, 25, 41–67.
- Cohen-Mansfield, J. and Perach, R.** (2015). Interventions for alleviating loneliness among older persons: a critical review. *American Journal of Health Promotion*, 29, e109–e125.
- Critical Appraisal Skills Programme (CASP)** (2017). *CASP Qualitative Checklist*. Available at: http://docs.wixstatic.com/ugd/dded87_25658615020e427da194a325e7773d42.pdf; last accessed 12 February 2018.
- Davies, N., Crowe, M. and Whitehead, L.** (2016). Establishing routines to cope with the loneliness associated with widowhood: a narrative analysis. *Journal of Psychiatric and Mental Health Nursing*, 23, 532–539.
- de Jong-Gierveld, J.** (1987). Developing and testing a model of loneliness. *Journal of Personality and Social Psychology*, 53, 119.
- Dickens, A., Richards, S. H., Greaves, C. J. and Campbell, J. L.** (2011). Interventions targeting social isolation in older people: a systematic review. *BMC Public Health*, 11, 647.
- Dykstra, P. A.** (2009). Older adult loneliness: myths and realities. *European Journal of Ageing*, 6, 91–100. doi:10.1007/s10433-009-0110-3.
- Elder, Jr., G. H., and Johnson, M. K.** (2003). The life course and aging: Challenges, lessons, and new directions. In R. A. Settersten (Ed.), *Invitation to the life course. Toward New Understandings of Later Life* (pp. 49–81). Amityville, NY: Baywood.
- Findlay, R. A.** (2003). Interventions to reduce social isolation amongst older people: where is the evidence? *Ageing & Society*, 23, 5647–5658.
- Gardiner, C., Geldenhuys, G. and Gott, M.** (2016). Interventions to reduce social isolation and loneliness among older people: an integrative review. *Health and Social Care in the Community*, doi:10.1111/hsc.12367.
- Graneheim, U. H. and Lundman, B.** (2010). Experiences of loneliness among the very old: the Umeå 85+ project. *Aging & Mental Health*, 14, 433–438.
- Hagan, R., Manktelow, R., Taylor, B. J. and Mallett, J.** (2014). Reducing loneliness amongst older people: a systematic search and narrative review. *Aging and Mental Health*, 18, 683–693.
- Hauge, S. and Kirkevold, M.** (2012). Variations in older persons' descriptions of the burden of loneliness. *Scandinavian Journal of Caring Sciences*, 26, 553–560.
- Holt-Lunstad, J., Smith, T. B. and Layton, J. B.** (2010). Social relationships and mortality risk: a meta-analytic

- review. *PLoS Medicine*, 7, e1000316. Available at: <https://doi.org/10.1371/journal.pmed.1000316>.
- Honigh-de Vlaming, R., Haveman-Nies, A., Bos-Oude Groeniger, I., de Groot, L. and van 't Veer, P.** (2014). Determinants of trends in loneliness among dutch older people over the period 2005–2010. *Journal of Aging and Health*, 26, 422–440. Available at: <https://doi.org/10.1177/0898264313518066>.
- Kharicha, K. et al.** (2017). What do older people experiencing loneliness think about primary care or community based interventions to reduce loneliness? A qualitative study in England. *Health and Social Care in the Community*, 25, 1733–1742. doi:10.1111/hsc.12438.
- Kirkevold, M., Moyle, W., Wilkinson, C., Meyer, J. and Hauge, S.** (2013). Facing the challenge of adapting to a life 'alone' in old age: the influence of losses. *Journal of Advanced Nursing*, 69, 394–403.
- Lou, V. W. Q. and Ng, J. W.** (2012). Chinese older adults' resilience to the loneliness of living alone: a qualitative study. *Aging & Mental Health*, 16, 1039–1046. doi:10.1080/13607863.2012.692764.
- Lunaigh, C. Ó. and Lawlor, B. A.** (2008). Loneliness and the health of older people. *International Journal of Geriatric Psychiatry*, 23, 1213–1221. doi:10.1002/gps.2054.
- Mental Health Foundation** (2010). *The Lonely Society*. London: The Mental Health Foundation.
- Peplau, L. A. and Perlman, D.** (1982). *Perspectives on Loneliness*. In L. A. Peplau and D. Perlman (Eds.), *Loneliness: A Sourcebook of Current Theory, Research and Therapy* (pp. 1–20). Chichester: John Wiley and Sons.
- Pettigrew, S. and Roberts, M.** (2008). Addressing loneliness in later life. *Aging & Mental Health*, 12, 302–309.
- Popay, J. et al.** (2006). *Guidance on the Conduct of Narrative Synthesis in Systematic Reviews: A Product from the ESRC Methods Programme* (Version 1). Lancaster: Lancaster University.
- Roos, V. and Klopper, H.** (2010). Older persons' experiences of loneliness: a South African perspective. *Journal of Psychology in Africa*, 20, 281–289.
- Smith, J. M.** (2012). Toward a better understanding of loneliness in community-dwelling older adults. *The Journal of Psychology*, 146, 293–311. doi:10.1080/00223980.2011.602132.
- Stanley, M., Moyle, W., Ballantyne, A., Jaworski, K., Corlis, M. and Oxlade, D.** (2010). 'Nowadays you don't even see your neighbours': loneliness in the everyday lives of older Australians. *Health & Social Care in the Community*, 18, 407–414. doi:10.1111/j.1365-2524.2010.00923.x.
- Strauss, A. and Corbin, J.** (2008). *Basics of Qualitative Research: Techniques and Procedures for Developing Grounded Theory*. Newbury Park, CA: Sage Publications.
- Sullivan, M. P., Victor, C. R. and Thomas, M.** (2016). Understanding and alleviating loneliness in later life: perspectives of older people. *Quality in Ageing and Older Adults*, 17, 168–178.
- Taube, E., Jakobsson, U., Midlov, P. and Kristensson, J.** (2016). Being in a bubble: the experience of loneliness among frail older people. *Journal of Advanced Nursing* 72, 631–640. doi:10.1111/jan.12853.
- Tong, A., Flemming, K., McInnes, E., Oliver, S. and Craig, J.** (2012). Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ. *BMC Medical Research Methodology*, 12, 181. Available at: <https://doi.org/10.1186/1471-2288-12-181>.
- Tong, A., Sainsbury, P. and Craig, J.** (2007). Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*, 19, 349–357.
- Valtorta, N. K., Kanaan, M., Gilbody, S., Ronzi, S. and Hanratty, B.** (2016). Loneliness and social isolation as risk factors for coronary heart disease and stroke: systematic review and meta-analysis of longitudinal observational studies. *Heart*, 102, 1009–1016.
- Victor, C., Scambler, S., Bowling, A. and Bond, J.** (2005). The prevalence of, and risk factors for, loneliness in later life: a survey of older people in Great Britain. *Ageing and Society*, 25, 357–375. doi:10.1017/S0144686X04003332.
- Victor, C., et al.** (2002). Has loneliness amongst older people increased? An investigation into variations between cohorts. *Ageing and Society*, 22, 585–597. doi:10.1017/S0144686X02008784.
- Weiss, R. S.** (1973). *Loneliness: The experience of emotional and social isolation*. Cambridge, MA: The MIT Press.
- Wilson, C. and Moulton, B.** (2010). Loneliness among older adults: a national survey of adults 45+. *Prepared by Knowledge Networks and Insight Policy Research*. Washington, DC: AARP.
- Windle, K., Francis, J. and Coomber, C.** (2011). *SCIE Research Briefing 39: Preventing Loneliness and Social Isolation: Interventions and Outcomes*. London: Social Care Institute for Excellence.

Appendix 1: Search terms

Older people

1. elder*.mp.
2. exp Geriatrics/
3. exp "Aged, 80 and over"/
4. old* person.mp.
5. old* people.mp.
6. exp Aging/ or ag?ing.mp.
7. old* age.mp.
8. senior*.mp.

Loneliness and social isolation

1. exp Loneliness/
2. lonel*.mp.
3. exp Social Isolation/
4. social isolation.mp.
5. solitude.mp.
6. solitary.mp.
7. liv* alone.mp.
8. exp Social Alienation/

Coping strategies

1. exp Self Care/
2. self manag*.mp.
3. exp Adaptation, Psychological/
4. (emotion* adj3 manag*).mp.
5. (feeling* adj3 manag*).mp.

6. (psycholog* adj3 manag*).mp.
7. (coping adj3 mechanism*).mp.
8. (psychological* adj3 adjust*).mp.
9. (emotion* adj3 adjust*).mp.
10. (behavioral* adj3 adjust*).mp.
11. (psychological* adj3 adapt*).mp.
12. psychological adjust*.mp
13. (psychological adj3 strat*).mp.
14. (emotion* adj3 strat*).mp.
15. exp Self Efficacy/
16. (coping adj3 strat*).mp.
17. (coping adj3 behavior*).mp.
18. exp Coping/
19. coping skill*.mp
20. self reliance.mp.
21. exp Resilience, Psychological/
22. resilience.mp.
23. manag* lonel*.mp
24. exp adaptive behavior/
25. exp coping behavior/

REVIEW

Remote delivery of psychological interventions for Parkinson's disease

Caitlin Swalwell,^{1,2} Nancy A. Pachana¹ and Nadeeka N. Dissanayaka^{1,2,3}

¹School of Psychology, The University of Queensland, Brisbane, Australia

²UQ Centre for Clinical Research, Faculty of Medicine, The University of Queensland, Brisbane, Australia

³Department of Neurology, Royal Brisbane & Woman's Hospital, Brisbane, Australia

ABSTRACT

Objective: Over two-thirds of Parkinson's disease (PD) patients experience comorbid neuropsychiatric symptoms, which adversely impact their quality of life and often require intervention. There is a preference for non-pharmacological, psychological approaches in addressing these symptoms. Given mobility limitations, travel burden, and cost, accessibility to psychological treatment can be problematic in this population. There has been a recent shift toward delivering care via telehealth in PD. Accordingly, this review aimed to examine remotely delivered psychological interventions for PD patients.

Results: Most of the telehealth studies for PD involved Cognitive Behavioral Therapy (CBT) based anxiety and depression telephone interventions with relatively short (one month) follow-up periods.

Conclusion: Although a preliminary work indicates efficacy, future studies should demonstrate the non-inferiority of these telehealth programs compared to face-to-face delivery, and examine the long-term outcomes of remotely delivered therapy. Video-conferencing (VC) appears to be a promising modality to overcome noted limitations of telephone delivery, and has demonstrated efficacy for PD speech programs. Further research should be conducted evaluating telehealth VC modalities for delivery of psychotherapy including CBT, as well as mindfulness-based therapy and acceptance and commitment therapy for remote treatment of depression and anxiety in PD.

Key words: Parkinson's disease, telehealth, depression, anxiety, cognitive behavioral therapy

Introduction

Parkinson's disease (PD) is a common neurodegenerative disorder. While it is primarily associated with progressive movement deterioration, debilitating non-motor symptoms are prevalent (Aarsland *et al.*, 1999). Neuropsychiatric symptoms, observed in more than 70% of PD sufferers (Riedel *et al.*, 2010), are increasingly recognized as important contributors to patient disability (Weintraub *et al.*, 2004), quality of life (Schrag *et al.*, 2000; Schrag, 2006; Dissanayaka *et al.*, 2010; Martinez-Martin *et al.*, 2011; Jiang *et al.*, 2013), and mortality (Hughes *et al.*, 2004). In fact, anxiety, which is experienced in an average of 31% of PD patients (Broen *et al.*, 2016), and depression, observed

in 35% of patients (Reijnders *et al.*, 2008), are thought to be the strongest PD quality of life predictors (Rahman *et al.*, 2008; Quelhas and Costa, 2009; Menon *et al.*, 2015). Evidence for the effectiveness of pharmacological interventions in treating PD neuropsychiatric symptoms is mixed (Price *et al.*, 2011; Troeung *et al.*, 2013), and concerns regarding unfavorable side effects, including exacerbation of motor problems in patients with complex medication regimes, have been noted (Rabinstein and Shulman, 2000; Veazey *et al.*, 2005; Chen and Marsh, 2014). Furthermore, impulse-control disorder (ICD) development in PD patients has been associated with dopamine agonist pharmacotherapy (Weintraub *et al.*, 2006). Consequently, non-pharmacological, psychological interventions provide a viable treatment alternative. Many PD patients, however, do not receive mental health treatment (Frisina *et al.*, 2008; Dobkin *et al.*, 2013), given their significant mobility impairments, a lack of available services locally (and a high prevalence of rural and remote PD patients), and

Correspondence should be addressed to: Nadeeka N. Dissanayaka, UQ Centre for Clinical Research, Royal Brisbane & Woman's Hospital, 71/918 Herston, Brisbane, QLD 4029, Australia. Phone: +61-7-33465577; Fax: +61-7-33465599. Email: n.dissanayaka@uq.edu.au. Received 25 Sep 2017; revision requested 7 Nov 2017; revised version received 5 Feb 2018; accepted 22 Feb 2018. First published online 11 May 2018.

transportation burden including distance traveled and costs (Dobkin *et al.*, 2013).

The success of face-to-face cognitive behavioral therapy (CBT) in addressing comorbid depression and/or anxiety (Feeney *et al.*, 2005; Dobkin *et al.*, 2007; Dobkin *et al.*, 2011a; Okai *et al.*, 2013; Troeung *et al.*, 2014; Shinmei *et al.*, 2016) and ICDs (Okai *et al.*, 2013) in PD patients has triggered a recent shift toward improving psychotherapy accessibility through the use of telehealth-based delivery in this group, thus enhancing patient-centered care. PD patients report specific interest in care delivered via home-based telehealth interventions (Dobkin *et al.*, 2013; Lubomski *et al.*, 2013). Pilot studies involving telehealth-based specialist care delivery through “virtual house calls” have demonstrated feasibility and considerable savings in travel distance and time for PD patients (Dorsey *et al.*, 2013), and large randomized-controlled trials (RCT) are underway (Achey *et al.*, 2014). Furthermore, remotely delivered assessments for PD patients in care facilities have demonstrated high patient and family satisfaction in addition to cost-effectiveness compared to transporting patients for face-to-face neurologist appointments (Barbour *et al.*, 2016).

To our knowledge, no previous review has specifically examined the remote delivery of psychological interventions in PD. This review aims to provide an update of the telehealth literature in this regard, and identify future directions for remote delivery of psychotherapy in this population. Given that most work has evaluated PD-specific anxiety and/or depression treatment, this review gives deliberate weight to these topics.

Methods

Major electronic databases (PubMed, PsycInfo, and Web of Science) were searched using the terms *Parkinson’s disease, telehealth, telephone, video-conferencing, ehealth, psychotherapy, cognitive behavioral therapy, acceptance and commitment therapy, and mindfulness*. Specified study inclusion criteria were as follows: (1) published in English, and (2) in a peer-reviewed journal; (3) conducted to evaluate the effect of a detailed psychological treatment as the primary intervention for PD patients; and, (4) the majority of the psychological treatment was delivered remotely (e.g. the initial and/or final session may be delivered in-person). Given the paucity of literature utilizing remote delivery, all trials (including uncontrolled pilot studies) published in December 2016 were considered eligible for inclusion. Abstracts were examined, and studies were excluded if psychiatric symptoms

were included as secondary outcomes for (1) non-psychotherapy-based interventions or (2) remote assessment/monitoring studies without treatment. Additionally, studies focused on evaluating remote care delivery across a range of subspecialties and/or conditions were excluded. References of identified articles were checked for additional citations.

Psychotherapy for anxiety and depression

A summary of study characteristics for telehealth-based psychological interventions for anxiety and depression in PD is provided in Table 1. To date, only five such studies have been published: Three of these involved telephone-delivered CBT (Veazey *et al.*, 2009; Dobkin *et al.*, 2011b; Calleo *et al.*, 2015), one involved a hybrid of CBT-based bibliotherapy and telephone support (Lawson *et al.*, 2013), and one involved internet-administered CBT (in the form of self-paced, online modules) with supplementary telephone support (Kraepelien *et al.*, 2015). All were pilot studies, and targeted comorbid depression and/or anxiety symptoms in older PD patients through one-on-one CBT.

Telephone-administered CBT

Telephone-administered CBT designed to simulate face-to-face treatment appears to be the most trialed remote delivery method for psychological interventions in this population. In more recent trials, CBT has been adapted to incorporate PD-specific symptom management and include caregivers (Dobkin *et al.*, 2011b; Calleo *et al.*, 2015), and also allows patients to specifically tailor certain aspects of skill training (Calleo *et al.*, 2015). Results are promising but variable, with significant improvements reported for anxiety only (Veazey *et al.*, 2009), depression only (Calleo *et al.*, 2015), or both depression and anxiety (Dobkin *et al.*, 2011b). However, the lack of a control group in Dobkin *et al.* (2011b) and the pilot nature of all studies (and consequential small sample sizes) likely limit statistical power in evaluation of depression and anxiety outcomes. For example, the between-group effect sizes for change scores at post-intervention and one-month follow-up for both anxiety and depression were large in Calleo *et al.* (2015) despite only the post-intervention reduction in depression reaching statistical significance (post-treatment: $d = 1.49$ for depression and 1.44 for anxiety; one-month follow-up: $d = 0.73$ for depression and 1.24 for anxiety).

There are limitations with these studies. Since all three limited follow-up periods to one month,

Table 1 Summary of studies involving telehealth-based CBT interventions for primary anxiety and/or depression in PD

| STUDY | DESIGN | SAMPLE CHARACTERISTICS: N (M, F), AGE, YEARS SINCE DIAGNOSIS, FINAL N COMPLETED ALL MEASURES | TARGET SYMPTOMS (ASSOCIATED INCLUSION CRITERIA) | INTERVENTION AND TELEHEALTH MODALITY; DURATION | CONTROL | PSYCHIATRIC SYMPTOM ± TELEHEALTH FINDINGS |
|------------------------------|--|--|--|--|--|--|
| Veazey <i>et al.</i> (2009) | Pilot, RCT (+ case example) data analyzed in completers | n = 10 (10, 0), 70.5 ± 8.22 years, NR, n = 7 | Anxiety (BAI ≥ 16) and/or Depression (PHQ-9 ≥ 10) | Telephone-administered, individual CBT; 8 × weekly sessions (following 1 × initial in-person session) | Supportive weekly telephone calls, 8 weeks | <ul style="list-style-type: none"> • Significant reduction in anxiety (BAI) for CBT group post-intervention, maintained at 1-month follow-up • Pre-treatment (CBT: M = 23.6, SD = 8.3; support: M = 23.8, SD = 9.4, $\eta^2 = 0.00$) • Post-treatment (CBT: M = 16.5, SD = 7.6; support: M = 21.3, SD = 11.3 $\eta^2 = 0.08$) • 1-month (CBT: M = 11, SD = 3.6; support: M = 21, SD = 7.8, $\eta^2 = 0.5$) • No difference in reduction in depression (PHQ-9) between groups • No reported measurements of patient satisfaction with telehealth; informal patient feedback indicated overall preference for telephone appointments • Significant reduction in depression (HAM-D 17, and BDI), anxiety (HAM-A), and negative thoughts (IQ), and improvement in coping (Brief Copie) following CBT |
| Dobkin <i>et al.</i> (2011b) | Pilot (pre-post comparison), uncontrolled Intent-to-treat analysis | n = 21 (8, 13), 65.86 ± 9.38 years, 7.45 ± 9.38 years, n = 20 | Depression (depressive disorder confirmed via SCID) NB. 24% of patients had secondary anxiety disorder diagnosis | Telephone-administered, individual CBT (+ printed supporting manual); 10 × weekly 60–90 minutes sessions | None | <ul style="list-style-type: none"> • Baseline: M = 21.29, SD = 6.48; midpoint: M = 15.33, SD = 6.63; endpoint: M = 13.38, SD = 6.63; follow-up: M = 13.84, 6.77, F = 18.49, p < 0.001, d = 1.21 • BDI • Baseline: M = 22.62, SD = 9.24; midpoint: M = 15.07, SD = 9.55; endpoint: M = 12.00, SD = 9.57, follow-up: M = 11.89, SD = 9.73, F = 23.68, p < 0.001, d = 1.13 • HAM-A • Baseline: M = 19.86, SD = 5.79; midpoint: M = 15.46, SD = 5.91; endpoint: M = 13.51, SD = 5.91; follow-up: M = 14.07, SD = 6.03, F = 15.59, p < 0.001, d = 1.09 |

Table 1 Continued

| STUDY | DESIGN | SAMPLE CHARACTERISTICS: N (M, F), AGE, YEARS SINCE DIAGNOSIS, FINAL N COMPLETED ALL MEASURES | TARGET SYMPTOMS (ASSOCIATED INCLUSION CRITERIA) | INTERVENTION AND TELEHEALTH MODALITY; DURATION | CONTROL | PSYCHIATRIC SYMPTOM ± TELEHEALTH FINDINGS |
|-------|--------|--|---|--|---------|---|
| | | | | | | <p><i>Negative thoughts (IQ)</i></p> <ul style="list-style-type: none"> ○ Baseline: $M = 7.33$, $SD = 4.62$; midpoint: $M = 6.74$, $SD = 4.83$; endpoint: $M = 5.33$, $SD = 4.83$; follow-up: $M = 4.28$, $SD = 5.08$, $F = 4.05$, $p = 0.01$, $d = 0.42$ <p><i>Brief COPE – positive reframing subscale</i></p> <ul style="list-style-type: none"> ○ Baseline: $M = 3.85$, $SD = 2.18$; midpoint: $M = 5.01$, $SD = 2.29$; endpoint: $M = 5.78$, $SD = 2.23$; follow-up: $M = 5.49$, $SD = 2.29$, $F = 7.09$, $p < 0.001$, $d = 0.88$ <p><i>Brief COPE – problem-focused subscale</i></p> <ul style="list-style-type: none"> ○ Baseline: $M = 15.50$, $SD = 5.79$; midpoint: $M = 16.56$, $SD = 6.06$; endpoint: $M = 18.64$, $SD = 5.94$; follow-up: $M = 16.90$, $SD = 6.06$, $F = 3.00$, $p = 0.04$, $d = 0.54$ <ul style="list-style-type: none"> • High perceived empathy from therapists during phone calls • Phone-based intervention rated “very helpful” |

Table 1 Continued

| STUDY | DESIGN | SAMPLE CHARACTERISTICS: N (M, F), AGE, YEARS SINCE DIAGNOSIS, FINAL N COMPLETED ALL MEASURES | TARGET SYMPTOMS (ASSOCIATED INCLUSION CRITERIA) | INTERVENTION AND TELEHEALTH MODALITY; DURATION | CONTROL | PSYCHIATRIC SYMPTOM ± TELEHEALTH FINDINGS |
|---------------------------------|--|---|---|--|---|---|
| Lawson <i>et al.</i> (2013) | Pilot, RCT data analyzed in completers | n = 54, (14, 18), 65.9 ± 8.3 years, 5.16 ± 3.95 years, n = 32 | Anxiety (HADS-A > 8) | Guided reading program (CBT-based, printed, self-help book with fortnightly telephone support) derived from <i>What? Me Worry?!</i> (an online self-help resource); 8 × self-paced chapters, 8 weeks | Printed information about worry only (first 2 × chapters of intervention program); 1 × telephone call, no management advice | <ul style="list-style-type: none"> • Significant increase in worry (PSWQ) from baseline to follow-up group for controls, and reduction in worry and intolerance of uncertainty (IUS) for intervention group • PSWQ <ul style="list-style-type: none"> ◦ Guided reading intervention (baseline: M = 48.82, SD = 15.97; 3-months: M = 41.88, SD = 18.00, (Z = -2.35, p = 0.019) ◦ Controls (baseline: M = 49.00, SD = 13.58; 3-months: M = 52.40, SD = 13.51, Z = -2.19, p = 0.028) • IUS <ul style="list-style-type: none"> ◦ Guided reading intervention (baseline: M = 65.76, SD = 20.44; 3-months: M = 54.71, SD = 18.93, Z = -1.96, p = 0.049) • No difference between groups in worry reduction • No reported measurements of patient satisfaction with telehealth; Informal feedback indicated some patients prefer face-to-face support • Significant reduction in HADS total, and depression (HADS-D) <ul style="list-style-type: none"> • <i>HADS total</i> <ul style="list-style-type: none"> ◦ Pre: M = 20.1, SD = 6.2; post: M = 15.3, SD = 6.0, t(8) = 3.09, p < 0.05, d = 0.79 • <i>HADS-D</i> <ul style="list-style-type: none"> ◦ Pre: M = 10.6, SD = 2.7; post: M = 7.8, SD = 2.5, t(8) = 2.44, p < 0.05, d = 1.08 • No change in anxiety (HADS-A) • Moderate overall patient satisfaction with intervention (CSQ-8, quantitative) • High satisfaction with telephone-based component (CSQ-8, qualitative) |
| Kraepelien <i>et al.</i> (2015) | Pilot (pre-post comparison), uncontrolled Intent-to-treat analysis | n = 9 (6, 3), 66.0 ± 11.6 years, 8.1 ± 3.9 years, n = 6 | Anxiety (HADS-A > 7) or Depression (HADS-D > 7) | Internet-based CBT (+ telephone support when needed); 11 x self-paced online modules (maximum), 12 weeks | None | <ul style="list-style-type: none"> • Significant reduction in HADS total, and depression (HADS-D) <ul style="list-style-type: none"> • <i>HADS total</i> <ul style="list-style-type: none"> ◦ Pre: M = 20.1, SD = 6.2; post: M = 15.3, SD = 6.0, t(8) = 3.09, p < 0.05, d = 0.79 • <i>HADS-D</i> <ul style="list-style-type: none"> ◦ Pre: M = 10.6, SD = 2.7; post: M = 7.8, SD = 2.5, t(8) = 2.44, p < 0.05, d = 1.08 • No change in anxiety (HADS-A) • Moderate overall patient satisfaction with intervention (CSQ-8, quantitative) • High satisfaction with telephone-based component (CSQ-8, qualitative) |

Table 1 Continued

| STUDY | DESIGN | SAMPLE CHARACTERISTICS: N (M, F), AGE, YEARS SINCE DIAGNOSIS, FINAL N COMPLETED ALL MEASURES | TARGET SYMPTOMS (ASSOCIATED INCLUSION CRITERIA) | INTERVENTION AND TELEHEALTH MODALITY; DURATION | CONTROL | PSYCHIATRIC SYMPTOM ± TELEHEALTH FINDINGS |
|----------------------|--|---|--|--|---|--|
| Calleo et al. (2015) | Pilot, controlled trial Data analyzed in completers | n = 16 (14, 2), 62.9 ± 7.3 years, NR, n = 11 | Anxiety (HADS-A > 4) and/or Depression (GDS-15 > 5), confirmed by SCID | Face-to-face (33%) or telephone-administered (67%) CBT (according to patient preference); 8 × 30–40 minutes sessions (maximum), 12 weeks | Usual care + mailed supplementary PD symptom education materials from National Parkinson's Foundation | <ul style="list-style-type: none"> • Greater reduction in depression (SIGH-D) from baseline to post-intervention (but not to 1-month follow-up) for CBT group <i>SIGH-D</i> <ul style="list-style-type: none"> ◦ CBT (baseline: M = 17.29, SD = 8.30; post-intervention, M = 12.14, SD = 7.84) vs usual care (baseline: M = 11.50, SD = 5.69; post-intervention: M = 13.75, SD = 3.10) ◦ Between-group ((mean difference for CBT = -5.14 (5.49), for usual care = 2.25 (4.79)), p = 0.045, d = 1.49) • No differences between groups in anxiety reduction (<i>SIGH-A</i>) • Large between-group effect sizes for anxiety and depression (at baseline and 1-month follow-up) <i>SIGH-A</i> <ul style="list-style-type: none"> ◦ CBT vs Usual Care (Mean difference for CBT = -8.17 (4.71), for usual care = -2.20 (5.97)), p = 0.10, d = 1.24) • <i>SIGH-D</i> <ul style="list-style-type: none"> ◦ CBT vs Usual Care (Mean difference for CBT = -5.00 (8.32), for usual care = 0.00 (6.60)), p = 0.52, d = 1.24 • Telephone-based delivery rated by patients as moderately effective |

NB. Only findings specific to psychiatric symptoms and telehealth delivery efficacy are reported. NR, not reported; BAI, Beck Anxiety Inventory; PHQ-9, Patient Health Questionnaire-9; HAM-D 17, Hamilton Rating Scale for Depression; SCID, Structured Clinical Interview for *Diagnostic and Statistical Manual*, Fourth Edition; BDI, Beck Depression Inventory; HAM-A, Hamilton Anxiety Rating Scale; IQ, Inference Questionnaire; HADS-A, Hospital Anxiety and Depression Scale Anxiety Score; HADS-D, Hospital Anxiety and Depression Scale Depression score; PSWQ, Penn State Worry Questionnaire; IUS, Intolerance of Uncertainty Scale; CSQ-8, Client Satisfaction Questionnaire; GDS-15, Geriatric Depression Scale-15; SIGH-D, Structured Interview Guide for the Hamilton Depression Scale; SIGH-A, Structured Interview Guide for the Hamilton Anxiety Scale.

the long-term effects of telephone-administered treatment are unknown. Of the 54 potential study participants with clinically significant anxiety and/or depression (based on DSM criteria) identified by Veazey *et al.* (2009) from neurology clinic outpatients, only 14 undertook baseline assessment, and of these only 10 of these met subsequent inclusion criteria, and 7 completed treatment. Thus, this study demonstrates that reduced interest, attrition and, non-completion can be problematic in this population, reducing sample sizes and limiting treatment effectiveness evaluation. Conversely, the higher retention rates for Dobkin *et al.* (2011b) (95%) and Calleo *et al.* (2015) (69%) may be linked to caregiver inclusion in treatment; that is, caregivers were offered their own supplementary telephone-based educational sessions to encourage patient support during treatment. Caregiver participation, however, may be linked to patient improvement.

Unlike Dobkin *et al.* (2011b) and Veazey *et al.* (2009), patients randomized to the CBT group in Calleo *et al.* (2015) elected whether to receive their treatment via face-to-face or telephone sessions. This was offered to maximize accessibility and flexibility in delivery, and two-thirds of sessions were delivered via telephone, allowing self-selection of delivery modality limits independent evaluation of telephone delivery efficacy.

Patient perceptions of telehealth effectiveness were formally assessed in only two of these studies; however, PD patients appear receptive to the modality. In Dobkin *et al.* (2011b), patients reported high perceived empathy from the phone therapist and rated the treatment as “very helpful,” and in Calleo *et al.* (2015) telephone delivery was rated as “moderately effective” with feedback indicating benefits regarding convenience, and reduced travel concerns and cost. However, some limitations were noted, including being unable to see the clinician and preferring in-person delivery for some specific skills (e.g. emotion management) (Calleo *et al.*, 2015). Additionally, informal patient feedback in Veazey *et al.* (2009) indicated an overall preference for telephone appointments by increasing scheduling flexibility and eliminating weekly travel burden, but the authors did note some difficulties detecting patient emotion. Thus, impaired patient–clinician emotional communication appears to be a common limitation in telephone-based delivery. This could be addressed through upgrading telecommunication to live video-conferencing (VC). To our knowledge, no prior studies have employed VC for psychological intervention delivery in this patient population. Notably, however, VC has been implemented effectively in the treatment of other

PD comorbidities; for example, Lee Silverman Voice Treatment (LSVT) delivered via VC for PD-related speech and language difficulties has demonstrated non-inferiority compared to face-to-face delivery (Constantinescu *et al.*, 2011; Theodoros *et al.*, 2016) and has been well-received by patients (Constantinescu *et al.*, 2010, 2011; Dias *et al.*, 2016).

Dobkin *et al.* (2011a, 2014) noted an association between improvements in mood and gains in executive functioning and verbal memory for depressed PD patients following face-to-face CBT. Given that their telephone-administered CBT pilot study also revealed significant improvements in PD patient mood (Dobkin *et al.*, 2011b) (post-treatment effect size for depression: $d = 1.21$ and for anxiety $d = 1.09$), a subsequent brief publication explored the neuropsychological outcomes of this treatment (Dobkin, 2014). Their findings indicate that (1) gains in verbal memory were associated with phone-based CBT delivery and (2) baseline working and verbal memory predicted depression improvement post-CBT. Overall, Dobkin (2014) suggests that the intact working memory is necessary for PD patients to garner optimal CBT benefit in general, whereas verbal memory is needed to yield a positive response to telephone-based intervention delivery specifically. Importantly, this suggests that the telephone-based psychological interventions necessitate higher order functioning, and are thus more suited to PD patients without extensive cognitive impairment.

Telephone-based CBT delivery for anxiety and depression in PD is feasible and shows preliminary efficacy both for patient outcomes and service satisfaction; however, the lack of large randomized-controlled trials limits conclusions. Caregiver involvement in treatment may increase retention rates, and intact verbal memory appears to be important to maximize treatment efficacy. VC could address noted modality limitations.

Beyond pure telephone-administered psychotherapy

Aforementioned telephone-based CBT interventions for anxiety and depression effectively mirror in-person delivery (in terms of content and duration of therapist contact). Given that the milder symptoms may negate the need for intensive psychotherapy, and concerns regarding a lack of available CBT-trained therapists to deliver treatment to all those who could benefit from it (Kraepelien *et al.*, 2015), some researchers have examined the feasibility of more patient-directed,

self-paced telehealth approaches to administer CBT in PD.

Lawson *et al.* (2013) compared a hybrid of bibliotherapy (including CBT-based self-guided exercises) with fortnightly telephone support to information only in anxious PD patients. Although the intervention group demonstrated a significant reduction in worry from baseline to three-month follow-up (baseline: $M = 48.82$, $SD = 15.97$; three-month follow-up: $M = 41.88$, $SD = 18.00$, $Z = -2.35$, $p = 0.019$), there was no significant difference between groups in reduction in worry. Notably, treatment compliance was not examined and high rates of attrition and failure to complete all outcome measures reduced the final sample to only 32 participants (from 54). Self-directed programs assume a certain level of initiative and motivation. Even Veazey *et al.* (2009) noted the need for participant initiative in completing homework tasks between phone-based CBT sessions. Notably, only 20% of participants in the Lawson *et al.* (2013) intervention group attempted all self-directed exercises, and several indicated a preference for face-to-face over telephone-based contact (however, overall patient satisfaction was not reported). Importantly, no apathy assessment was conducted. Apathy is a common neuropsychiatric symptom in PD, affecting up to 40% of patients (den Brok *et al.*, 2015), which could limit the capacity for self-directed care.

Kraepelien *et al.* (2015) conducted the first uncontrolled pilot study examining the efficacy and feasibility of internet-based CBT (ICBT) with telephone support (as needed) to treat anxiety and depression in PD. A significant reduction in depression only was observed (mean difference = -2.8 (95% CI = -5.4 to -0.1 ; $d = 1.08$; $t(8) = 2.44$; $p < 0.05$); however, the small sample and the absence of a control group limit the evaluation of intervention efficacy. Patients reported moderate overall service satisfaction, with high satisfaction for the telephone-based component; however, many participants did not complete homework exercises. Additionally, one-third of patients prematurely ceased treatment. As with Lawson *et al.* (2013), this could indicate reduced motivation; however, no motivation or apathy measures were administered. Importantly, this study demonstrates reduced therapist requirements. Since most information was delivered through the internet platform, therapists spent less than 13 minutes in communication with each participant per week. However, patient feedback indicated a desire for greater therapist communication.

Overall, these self-directed programs offer even greater delivery flexibility and reduced therapist input. However, only a preliminary work has been

conducted, long-term effects are unknown, no study has involved PD caregivers, and the common presentation of co-morbid apathy in PD has been seemingly overlooked. This should be formally assessed in future studies. Importantly, this initial work highlights an apparent PD patient preference for regular, face-to-face therapist contact, even when mild depression and/or anxiety symptoms are the targets of treatment. In Lawson *et al.* (2013), VC (rather than telephone support) could help address requests for face-to-face therapist communication, while still maintaining remote delivery status. In Kraepelien *et al.* (2015), patient requests for greater therapist contact could be addressed by scheduling weekly VC sessions (rather than having patients seek telephone support on an “as needed” basis). Notably, however, this would be at the expense of reduced therapist workload—a primary advantage of self-directed treatment programs.

Other remotely delivered psychological interventions for PD

Most telehealth studies in PD have focused on the delivery of psychological interventions for anxiety and/or depression. As indicated above, apathy is being recognized as an increasingly important PD comorbidity, not necessarily associated with depression, which has been found to interfere with PD motor disability treatment, exacerbating disability, and lowering quality of life and ability to carry out activities of daily living (ADL) (Weintraub *et al.*, 2004; Benito-Leon *et al.*, 2012; Laatu *et al.*, 2013). Thus, its consideration and treatment are essential in this population. Recently, Butterfield *et al.* (2016) uncontrolled piloted the first study involving a PD-specific telehealth-based behavioral activation intervention targeting apathy, namely the Parkinson’s Active Living (PAL) program, in patients without dementia. This study is summarized in Table 2.

As the first study addressing PD-related apathy via telehealth, this six-week program shows promise, with significant improvements reported in psychiatric symptoms (apathy: $d = 0.77$; depression: $d = 0.70$), which were maintained for one month following program cessation. Notably, apathy levels decreased to normal limits for over a third of the sample. However, this brief follow-up period limits evaluation of long-term intervention effects, and the lack of a control group limits conclusions. Clearly, a large-scale RCT with a longer follow-up period is required. Patient satisfaction with the overall program and the delivered treatment and materials were high, and attrition was low

Table 2 Study characteristics of Butterfield *et al.* (2016)

| | SAMPLE CHARACTERISTICS: N (M, F), AGE, YEARS SINCE DIAGNOSIS | TARGET SYMPTOMS (ASSOCIATED INCLUSION CRITERIA) | INTERVENTION AND TELEHEALTH MODALITY; DURATION | SYMPTOM OUTCOMES AND TELEHEALTH-RELATED FINDINGS |
|--|--|---|---|---|
| STUDY DESIGN | 27 (22, 5), 66 ± 10.7 years, 10.1 ± 6.2 years | Apathy (AES > 35) | Weekly telephone support sessions (10–20 minutes) following initial face-to-face planning session to identify activity goals for different life domains, automated phone reminders prior to each planned activity + workbook; 6 weeks | <ul style="list-style-type: none"> • Significant reduction in apathy (AES) post-intervention, maintained at 1-month follow-up <ul style="list-style-type: none"> ○ Baseline: $M = 42.1$, $SD = 6.0$; ○ post-intervention: $M = 36.1$, $SD = 8.3$, follow-up: $M = 37.0$, $SD = 7.4$ ■ Baseline and post-intervention: $t(26) = -4.002$, $d = 0.77$ ■ Baseline and follow-up: $t(19) = -4.264$, $d = 0.95$ • Significant reduction in depression (GDS) at post-intervention, maintained at 1-month follow-up <ul style="list-style-type: none"> ○ Baseline: $M = 13.61$, $SD = 7.04$; ○ post-intervention: $M = 10.78$, $SD = 6.54$, follow-up: $M = 37.0$, $SD = 7.4$ ■ Baseline and post-intervention: $t(22) = -3.380$, $d = 0.77$ ■ Baseline and follow-up: $t(15) = -4.002$, $d = 0.95$ • Significant increase in patient quality of life (PDQ-39) at post-intervention <ul style="list-style-type: none"> ○ Baseline: $M = 30.82$, $SD = 15.18$; ○ post-intervention: $M = 25.51$, $SD = 13.51$, $d = 0.50$, $t(23) = -2.458$ • Majority of participants reported satisfaction with the overall program (CSQ-8), and treatment and materials provided (PSQ-39) • 60% patient satisfaction with automated phone reminders; no reported measurements of patient satisfaction with telephone-based therapist support specifically |
| Pilot, uncontrolled (pre–post comparison) Data analyzed in completers | | | | |

AES, Apathy Evaluation Scale; GDS, Geriatric Depression Scale; PDQ-39, Parkinson's Disease Quality of Life Scale; CSQ-8, Client Satisfaction Questionnaire; PSQ-39, Participant Satisfaction Questionnaire.

(despite the absence of caregiver involvement in the intervention specifically), indicating program feasibility and receptiveness. Unfortunately, no explicit measure of patient satisfaction with the telephone support sessions was provided. However, the authors do indicate interest in upgrading from telephone to VC-based support delivery given its success in delivering treatment for other PD comorbidities (e.g. LSVT). The automated phone reminder element of the program received mixed support from patients, and was time-consuming for providers. As suggested by Butterfield *et al.* (2016), perhaps a more synchronized, modern approach (e.g. reminders distributed through text messages or a smart-phone application) could be feasible while still targeting self-initiative deficits.

Interestingly, verbal phonemic fluency correlated with apathy-specific response to treatment, suggesting the current PAL program necessitates intact executive function to garner greater benefit. This is an important consideration for PD given the high prevalence of dementia, and is consistent with the findings for CBT treatment benefit for depression in PD (Dobkin *et al.*, 2011b; Dobkin, 2014).

Future directions for telehealth in PD

Thus far, studies involving telehealth-based delivery of psychological interventions for PD patients are preliminary, and have primarily focused on treating depression with or without comorbid anxiety using CBT strategies. Furthermore, data handling approaches used in studies were mixed. While missing data were handled using intent-to-treat analysis by Dobkin *et al.* (2011b) and Kraepelien *et al.* (2015), others analyzed data only in completers. While reviewed studies demonstrate feasibility, there is clearly a need for large-scale, non-inferiority studies comparing the efficacy of telehealth delivered interventions to conventional face-to-face treatment, and for follow-up durations exceeding one month with appropriate data analysis approaches of handling missing data, in order for full treatment potential to be realized. Moreover, none of the studies reported whether a landline or a mobile device was used for telehealth-based delivery. Given the increasing number of people using mobile devices, the sound quality and reliability of connection can be compromised in wireless modality compared to landline connections. Therefore, it is important to specify the number of persons using landline or mobile phones in future reporting. Although most papers reviewed here have indicated at least moderate patient satisfaction with overall telephone-based service provision (or part thereof),

overcoming modality limitations reported by both patients and clinicians by adapting delivery to VC appears to be a common theme. The success of vocal telerehabilitation programs delivered via VC for PD could provide a guiding framework for clinicians intending to adapt telephone-delivered psychological interventions (Constantinescu *et al.*, 2010; 2011; Dias *et al.*, 2016; Theodoros *et al.*, 2016). It is essential to have an appropriate internet connection for VC-based interventions and this may limit access to VC in environments that there are less likely to have internet connections such as for individuals living in remote areas or with lower socio-economic status. Furthermore, allowing patients to provide open-ended feedback on service components for formal qualitative assessment in future trials, irrespective of the specific telehealth intervention, would be beneficial. In addition to participant satisfaction, it would be of interest to measure participants' level of concern about privacy with telehealth-based interventions in future. This may not be limited to the security of the line, but also whether the individual has a place at their residence where they feel they have sufficient privacy to engage in a verbal psychotherapy that might be overheard by others in the household.

It is important to note here that considerable symptom overlap between PD and the neuropsychiatric conditions discussed in this review has raised questions as to the suitability of established measurement tools in identifying and assessing PD-specific anxiety and depression symptoms. Recommendations have been provided (see Pachana *et al.*, 2013; Dissanayaka *et al.*, 2015 for reviews). Only one identified telehealth study in the present review by Calleo *et al.* (2015) employed a scale recommended for use with PD patients (the GDS-15); however, their assessment cut-off was not adjusted (up to seven as per recommendations) (Pachana *et al.*, 2013) for this clinical group. This suggests that the measures used in these studies (including the Beck Anxiety Inventory (BAI), Hamilton Anxiety Rating Scale (HAM-A), and Hospital Anxiety and Depression Scale Anxiety Score (HADS-A)) may have been inappropriate or incorrectly implemented for PD patients, and is an important consideration for future work evaluating the true efficacy of telehealth-based psychological interventions. Future anxiety interventions could utilize the Parkinson's Anxiety Scale and/or the Geriatric Anxiety Inventory for assessment, both of which have now been validated for use in PD patients (Dissanayaka *et al.*, 2015).

Beyond depression and anxiety, telehealth-based delivery of psychological interventions for PD comorbidities is lacking, despite success in

face-to-face interventions. In a waitlist-controlled trial for PD patients with associated ICDs, which are usually managed by adjusting PD medication (Zhang *et al.*, 2016), 12 weekly sessions of face-to-face CBT (complementing standard medical care) produced a significant reduction in ICD symptom severity and impact (Okai *et al.*, 2013). Secondary outcomes were significant reductions in depression and anxiety six months following treatment initiation. Despite this, no studies have delivered psychological interventions for PD-related ICDs via remote methods. In Okai *et al.* (2013) nurse-therapist visits enabled most CBT to be provided to patients within their own homes. Given this, adaptation to telehealth delivery could be feasible, especially with caregiver assistance.

To our knowledge, no study has delivered mindfulness-based or acceptance and commitment therapy (ACT) to PD patients using telehealth modalities. Preliminary face-to-face mindfulness interventions in PD have demonstrated short-term improvements in motor symptoms (Pickut *et al.*, 2015; Dissanayaka *et al.*, 2016a) and non-motor symptoms, including depression (Cash *et al.*, 2016; Dissanayaka *et al.*, 2016a), anxiety (Dissanayaka *et al.*, 2016a), cognition (Cash *et al.*, 2016; Dissanayaka *et al.*, 2016a), distress (Dissanayaka *et al.*, 2016a), and coping skills (Fitzpatrick *et al.*, 2010). A recent six-week group mindfulness intervention involving lifestyle management strategies for PD patients showed a significant reduction in depression and stress, and improvements in mindfulness and ADL at six-month follow-up (Advocat *et al.*, 2016). In-person ACT-based interventions for wearing-off related anxiety (Dissanayaka *et al.*, 2016b) associated with prolonged dopamine-replacement therapy in PD are currently underway (Ghielen *et al.*, 2015). These promising findings support the future adaptation of such psychological interventions for flexible, home-based delivery, thereby increasing telehealth-based treatment options for PD patients beyond CBT, and enhancing patient-centered care.

As the population ages, and PD prevalence increases, the need for PD-associated neuropsychiatric treatments will concurrently grow. The accessibility and availability of psychological interventions will become an increasingly important consideration, given mobility, transport and cost concerns for PD patients (Dobkin *et al.*, 2013). This review highlighted the infancy of telehealth-based psychological treatment provision in PD. There is a substantial literature gap regarding the development of telehealth interventions for PD-specific neuropsychiatric symptoms outside anxiety and depression. The PAL program (Butterfield *et al.*, 2016) appears to be the first intervention

developed to address this need. Furthermore, the non-inferiority and long-term outcomes of remotely delivered anxiety and depression treatments for this group are yet to be demonstrated.

Conflict of interest

None.

Description of authors' roles

C. Swalwell formulated the concept, organized and executed the review, and wrote the first draft. N.A. Pachana undertook the critical revision of the manuscript. N.N. Dissanayaka formulated the conception and organized the review, and also carried out the critical revision of drafts.

Acknowledgments

Dr N.N. Dissanayaka is supported by the Lions Medical Research Foundation fellowship.

References

- Aarstrand, D. *et al.* (1999). Range of neuropsychiatric disturbances in patients with Parkinson's disease. *Journal of Neurology, Neurosurgery, and Psychiatry*, 67, 492–496.
- Achey, M. A. *et al.* (2014). Virtual house calls for Parkinson disease (Connect.Parkinson): study protocol for a randomized, controlled trial. *Trials*, 15, 465.
- Advocat, J., Enticott, J., Vandenberg, B., Hased, C., Hester, J. and Russell, G. (2016). The effects of a mindfulness-based lifestyle program for adults with Parkinson's disease: a mixed methods, wait list controlled randomised control study. *BMC Neurology*, 16, 166.
- Barbour, P. J., Arroyo, J., High, S., Fichera, L. B., Staska-Pier, M. M. and McMahon, M. K. (2016). Telehealth for patients with Parkinson's disease: delivering efficient and sustainable long-term care. *Hospital Practice (1995)*, 44, 92–97.
- Benito-Leon, J., Cubo, E. and Coronell, C. (2012). Impact of apathy on health-related quality of life in recently diagnosed Parkinson's disease: the ANIMO study. *Movement Disorders*, 27, 211–218.
- Broen, M. P., Narayan, N. E., Kuijff, M. L., Dissanayaka, N. N. and Leentjens, A. F. (2016). Prevalence of anxiety in Parkinson's disease: A systematic review and meta-analysis. *Movement Disorders*, 31, 1125–1133.
- Butterfield, L. C., Cimino, C. R., Salazar, R., Sanchez-Ramos, J., Bowers, D. and Okun, M. S. (2016). The Parkinson's Active Living (PAL) Program: a behavioral intervention targeting apathy in Parkinson's disease. *Journal of Geriatric Psychiatry and Neurology*, 30, 11–25.
- Calleo, J. S. *et al.* (2015). A pilot study of a cognitive-behavioral treatment for anxiety and depression

- in patients with Parkinson disease. *Journal of Geriatric Psychiatry and Neurology*, 28, 210–217.
- Cash, T. V., Ekouevi, V. S., Kilbourn, C. and Lageman, S. K.** (2016). Pilot study of a mindfulness-based group intervention for individuals with Parkinson's disease and their caregivers. *Mindfulness*, 7, 361–371.
- Chen, J. J. and Marsh, L.** (2014). Anxiety in Parkinson's disease: identification and management. *Therapeutic Advances in Neurological Disorders*, 7, 52–59.
- Constantinescu, G. A., Theodoros, D. G., Russell, T. G., Ward, E. C., Wilson, S. J. and Wootton, R.** (2010). Home-based speech treatment for Parkinson's disease delivered remotely: a case report. *Journal of Telemedicine and Telecare*, 16, 100–104.
- Constantinescu, G. A., Theodoros, D. G., Russell, T. G., Ward, E. C., Wilson, S. J. and Wootton, R.** (2011). Treating disordered speech and voice in Parkinson's disease online: a randomized controlled non-inferiority trial. *International Journal of Language & Communication Disorders*, 46, 1–16.
- den Brok, M. G., van Dalen, J. W., van Gool, W. A., Moll van Charante, E. P., de Bie, R. M. and Richard, E.** (2015). Apathy in Parkinson's disease: a systematic review and meta-analysis. *Movement Disorders*, 30, 759–769.
- Dias, A. E., Limongi, J. C., Barbosa, E. R. and Hsing, W. T.** (2016). Voice telerehabilitation in Parkinson's disease. *CoDas*, 28, 176–181.
- Dissanayaka, N. N. et al.** (2010). Anxiety disorders in Parkinson's disease: prevalence and risk factors. *Movement Disorders*, 25, 838–845.
- Dissanayaka, N. N. et al.** (2016a). Mindfulness for motor and non-motor dysfunctions in Parkinson's disease. *Parkinson's Disease*, 2016, 7109052.
- Dissanayaka, N. N. et al.** (2016b). Disease-specific anxiety symptomatology in Parkinson's disease. *International Psychogeriatrics*, 28, 1153–1163.
- Dissanayaka, N. N., Torbey, E. and Pachana, N. A.** (2015). Anxiety rating scales in Parkinson's disease: a critical review updating recent literature. *International Psychogeriatrics*, 27, 1777–1784.
- Dobkin, R. D. et al.** (2011a). Cognitive behavioral therapy for depression in Parkinson's disease: a randomized, controlled trial. *The American Journal of Psychiatry*, 168, 1066–1074.
- Dobkin, R. D. et al.** (2011b). Telephone-based cognitive-behavioral therapy for depression in Parkinson disease. *Journal of Geriatric Psychiatry and Neurology*, 24, 206–214.
- Dobkin, R. D. et al.** (2014). Neuropsychological outcomes following psychosocial intervention for depression in Parkinson's disease. *The Journal of Neuropsychiatry and Clinical Neurosciences*, 26, 57–63.
- Dobkin, R. D.** (2014). The relationship between telephone-administered cognitive-behavioral therapy for depression and neuropsychological functioning in Parkinson's disease. *The Journal of Neuropsychiatry and Clinical Neurosciences*, 26, E10–11.
- Dobkin, R. D., Allen, L. A. and Menza, M.** (2007). Cognitive-behavioral therapy for depression in Parkinson's disease: a pilot study. *Movement Disorders*, 22, 946–952.
- Dobkin, R. D., Rubino, J. T., Friedman, J., Allen, L. A., Gara, M. A. and Menza, M.** (2013). Barriers to mental health care utilization in Parkinson's disease. *Journal of Geriatric Psychiatry and Neurology*, 26, 105–116.
- Dorsey, E. R. et al.** (2013). Randomized controlled clinical trial of “virtual house calls” for Parkinson disease. *JAMA Neurology*, 70, 565–570.
- Feeney, F., Egan, S. and Gasson, N.** (2005). Treatment of depression and anxiety in Parkinson's disease: a pilot study using group cognitive behavioral therapy. *Clinical Psychologist*, 9, 31–38.
- Fitzpatrick, L., Simpson, J. and Smith, A.** (2010). A qualitative analysis of mindfulness-based cognitive therapy (MBCT) in Parkinson's disease. *Psychology and Psychotherapy*, 83, 179–192.
- Frisina, P. G., Borod, J. C., Foldi, N. S. and Tenenbaum, H. R.** (2008). Depression in Parkinson's disease: health risks, etiology, and treatment options. *Neuropsychiatric Disease and Treatment*, 4, 81–91.
- Ghielen, I. et al.** (2015). BEWARE: Body awareness training in the treatment of wearing-off related anxiety in patients with Parkinson's disease: study protocol for a randomized controlled trial. *Trials*, 16, 283.
- Hughes, T. A., Ross, H. F., Mindham, R. H. and Spokes, E. G.** (2004). Mortality in Parkinson's disease and its association with dementia and depression. *Acta Neurologica Scandinavica*, 110, 118–123.
- Jiang, J. L., Tsai, S. T., Hsieh, T. C., Lee, C. W., Lin, S. H. and Chen, S. Y.** (2013). The impact of motor and depressive symptoms on quality of life in patients with Parkinson's disease. *Tzu Chi Medical Journal*, 25, 175–178.
- Kraepelien, M., Svenningsson, P., Lindfors, N. and Kaldo, V.** (2015). Internet-based cognitive behavioral therapy for depression and anxiety in Parkinson's disease—a pilot study. *Internet Interventions*, 2, 1–6.
- Laatu, S., Karrasch, M., Martikainen, K. and Marttila, R.** (2013). Apathy is associated with activities of daily living ability in Parkinson's disease. *Dementia and Geriatric Cognitive Disorders*, 35, 249–255.
- Lawson, R. A., Millar, D., Brown, R. G. and Burn, D. J.** (2013). Guided self-help for the management of worry in Parkinson's disease: a pilot study. *Journal of Parkinson's Disease*, 3, 61–68.
- Lubomski, M., Rushworth, R. L., Lee, W., Bertram, K. and Williams, D. R.** (2013). A cross-sectional study of clinical management, and provision of health services and their utilisation, by patients with Parkinson's disease in urban and regional Victoria. *Journal of Clinical Neuroscience*, 20, 102–106.
- Martinez-Martin, P., Rodriguez-Blazquez, C., Kurtis, M. M. and Chaudhuri, K. R.** (2011). The impact of non-motor symptoms on health-related quality of life of patients with Parkinson's disease. *Movement Disorders*, 26, 399–406.
- Menon, B. et al.** (2015). Parkinson's disease, depression, and quality-of-life. *Indian Journal of Psychological Medicine*, 37, 144–148.
- Okai, D. et al.** (2013). Trial of CBT for impulse control behaviors affecting Parkinson patients and their caregivers. *Neurology*, 80, 792–799.
- Pachana, N. A. et al.** (2013). Clinical issues in the treatment of anxiety and depression in older adults with Parkinson's disease. *Movement Disorders*, 28, 1930–1934.

- Pickut, B. et al.** (2015). Mindfulness training among individuals with Parkinson's disease: neurobehavioral effects. *Parkinson's Disease*, 2015, 816404.
- Price, A. et al.** (2011). Antidepressants for the treatment of depression in neurological disorders: a systematic review and meta-analysis of randomised controlled trials. *Journal of Neurology, Neurosurgery, and Psychiatry*, 82, 914–923.
- Quelhas, R. and Costa, M.** (2009). Anxiety, depression, and quality of life in Parkinson's disease. *The Journal of Neuropsychiatry and Clinical Neurosciences*, 21, 413–419.
- Rabinstein, A. A. and Shulman, L. M.** (2000). Management of behavioral and psychiatric problems in Parkinson's disease. *Parkinsonism & Related Disorders*, 7, 41–50.
- Rahman, S., Griffin, H. J., Quinn, N. P. and Jahanshahi, M.** (2008). Quality of life in Parkinson's disease: the relative importance of the symptoms. *Movement Disorders*, 23, 1428–1434.
- Reijnders, J. S., Ehrt, U., Weber, W. E., Aarsland, D. and Leentjens, A. F.** (2008). A systematic review of prevalence studies of depression in Parkinson's disease. *Movement Disorders*, 23, 183–189; quiz 313.
- Riedel, O. et al.** (2010). Frequency of dementia, depression, and other neuropsychiatric symptoms in 1,449 outpatients with Parkinson's disease. *Journal of Neurology*, 257, 1073–1082.
- Schrag, A.** (2006). Quality of life and depression in Parkinson's disease. *Journal of the Neurological Sciences*, 248, 151–157.
- Schrag, A., Jahanshahi, M. and Quinn, N.** (2000). What contributes to quality of life in patients with Parkinson's disease? *Journal of Neurology, Neurosurgery, and Psychiatry*, 69, 308–312.
- Shinmei, I. et al.** (2016). Cognitive behavioral therapy for depression in Japanese Parkinson's disease patients: a pilot study. *Neuropsychiatric Disease and Treatment*, 12, 1319–1331.
- Theodoros, D. G., Hill, A. J. and Russell, T. G.** (2016). Clinical and quality of life outcomes of speech treatment for Parkinson's disease delivered to the home via telerehabilitation: a non-inferiority randomized controlled trial. *American Journal of Speech-Language Pathology*, 25, 214–232.
- Troeng, L., Egan, S. J. and Gasson, N.** (2013). A meta-analysis of randomised placebo-controlled treatment trials for depression and anxiety in Parkinson's disease. *PLoS One*, 8, e79510.
- Troeng, L., Egan, S. J. and Gasson, N.** (2014). A waitlist-controlled trial of group cognitive behavioural therapy for depression and anxiety in Parkinson's disease. *BMC Psychiatry*, 14, 19.
- Veazey, C., Aki, S. O., Cook, K. F., Lai, E. C. and Kunik, M. E.** (2005). Prevalence and treatment of depression in Parkinson's disease. *The Journal of Neuropsychiatry and Clinical Neurosciences*, 17, 310–323.
- Veazey, C., Cook, K. F., Stanley, M., Lai, E. C. and Kunik, M. E.** (2009). Telephone-administered cognitive behavioral therapy: a case study of anxiety and depression in Parkinson's disease. *Journal of Clinical Psychology in Medical Settings*, 16, 243–253.
- Weintraub, D. et al.** (2006). Association of dopamine agonist use with impulse control disorders in Parkinson disease. *Archives of Neurology*, 63, 969–973.
- Weintraub, D., Moberg, P. J., Duda, J. E., Katz, I. R. and Stern, M. B.** (2004). Effect of psychiatric and other non-motor symptoms on disability in Parkinson's disease. *Journal of the American Geriatrics Society*, 52, 784–788.
- Zhang, S. et al.** (2016). Management of impulse control disorders in Parkinson's disease. *International Psychogeriatrics*, 28, 1597–1614.

REVIEW

Life story books for people with dementia: a systematic review

Teuntje R. Elfrink,¹ Sytse U. Zuidema,² Miriam Kunz² and Gerben J. Westerhof¹

¹Department of Psychology, Health and Technology, University of Twente, Enschede, the Netherlands

²Department of General Practice and Elderly Care Medicine, University of Groningen, University Medical Center Groningen, Groningen, the Netherlands

ABSTRACT

Objectives: There is an increasing evidence that reminiscence therapy is effective in improving cognitive functions and reducing depressive symptoms in people with dementia. Life story books (LSBs) are frequently used as a reminiscence tool to support recollecting autobiographical memories. As little is known about how LSBs are used and what type of studies have been employed to evaluate LSB interventions, we conducted a systematic review.

Methods: The electronic databases Scopus, PubMed, and PsychINFO as well as reference lists of existing studies were searched to select eligible articles. Out of the 55 studies found, 14 met the inclusion criterion of an original empirical study on LSBs in people with dementia.

Results: The majority of the LSBs were tangible books, although some digital applications were also found. The LSBs were created mostly in individual sessions in nursing homes with a median of six sessions. Some studies only focused on the person with dementia, while others also examined (in)formal caregivers. Most studies used qualitative interviews, case studies, and/or (pilot) randomized controlled trial (RCTs) with small sample sizes. Qualitative findings showed the value of LSBs in triggering memories and in improving the relation with the person with dementia. Quantitative effects were found on, e.g. autobiographical memory and depression of persons with dementia, quality of relationship with informal caregivers, burden of informal caregivers, and on attitudes and knowledge of formal caregivers.

Conclusions: This systematic review confirms that the use of LSBs to support reminiscence and person-centered care is promising, but larger RCTs or implementation studies are needed to establish the effects of LSBs on people with dementia.

Key words: life story book, reminiscence, dementia, systematic review, person-centered care, caregivers

Introduction

In 2015, the number of people living with dementia worldwide was estimated at 46.8 million. This number is expected to increase to 74.7 million in 2030 and 131.5 million in 2050 (Prince *et al.*, 2015). Dementia is often accompanied by neuropsychiatric symptoms (NPS). Dementia and the related NPS not only affect the quality of life of the person with dementia, but also result in a higher burden of informal caregivers and a lower quality of their lives (De Vugt *et al.*, 2003; Peeters *et al.*, 2012; Conde-Sala *et al.*, 2016). Dementia often leads to disability and a high and expanding need for care and support of a caregiver (Prince *et al.*,

2013). Overall, dementia is among the top five with the highest burden of disease for persons over 65 years and it belongs to the diseases with the highest burden for informal caregivers (McKeith and Cummings, 2005). This burden includes physical, emotional, and economic aspects (World Health Organization, 2017). Furthermore, dementia is one of the most costly diseases worldwide (Prince *et al.*, 2015; World Health Organization, 2017).

Higgs and Gilleard (2017) plead for a shift to a more person-centered approach in dementia care: person-centered care (PCC). The shift from medical, routine-driven care to personalized, individualized care – regardless of the cognitive or functional capacity of the patients – is called “the culture change” (Koren, 2010). In 2016, the American Geriatrics Society Expert Panel developed a summarizing definition of PCC: “*Person-centered care means that individuals’ values and preferences are elicited and, once expressed, guide*

Correspondence should be addressed to: Teuntje R. Elfrink, MSc, Department of Psychology, Health and Technology, University of Twente, P.O. Box 217, 7500 AE Enschede, the Netherlands. Email: t.r.elfrink@utwente.nl. Received 5 Dec 2017; revision requested 5 Feb 2018; revised version received 7 Feb 2018; accepted 26 Feb 2018. First published online 18 July 2018.

all aspects of their health care, supporting their realistic health and life goals. Person-centered care is achieved through a dynamic relationship among individuals, others who are important to them, and all relevant providers. This collaboration informs decision-making to the extent that the individual desires (AGS, 2016).

PCC can be enhanced by using a person's biography and memories (Clarke, 2000; Clarke *et al.*, 2003). The use of written or oral life histories to improve psychological well-being is described as reminiscence and is often used with people with dementia (McKeown *et al.*, 2006; VandenBos, 2006). Several (systematic) reviews have shown that reminiscence activities can contribute to the mental health and quality of life of persons with dementia (Woods *et al.*, 2005; Subramaniam and Woods, 2012; Blake, 2013). A recent meta-analysis of Huang and colleagues (2015) concluded that reminiscence therapy has a small effect on improving cognitive functioning and a moderate effect on reducing depressive symptoms in older persons with dementia, predominantly for those living in institutions.

The creation of a life story book (LSB) is a common approach in reminiscence (McKeown *et al.*, 2006). The use of LSBs with personal memories that are constructed together with the person with dementia is especially promising (Subramaniam and Woods, 2012). Besides the recollection of personal memories, receiving a tangible output in the form of a LSB is highly valued by patients (Morgan and Woods, 2010). In recent years, LSBs are used for multiple purposes and in multiple settings, while studies that evaluate LSB interventions for people with dementia are emerging. To provide an overview of how LSBs are used and what is known from research, we conducted a systematic review with two guiding questions:

1. How are LSBs used in dementia care?
2. What are the designs and findings of studies on the use of LSBs?

Methods

This systematic review was conducted according to the preferred reporting items for systematic reviews and meta-analyses statement (Moher *et al.*, 2015).

Inclusion and exclusion criteria

POPULATION

This review concerns people with mild cognitive impairment (MCI) or dementia.

TYPES OF INTERVENTIONS

Interventions that use LSBs were eligible for inclusion. There had to be a clear description of the LSB, and the LSB had to contain not only biographical facts but also autobiographical elements or memories.

OUTCOMES OF INTEREST

The outcomes of interest were (1) the use of LSBs and (2) the designs and findings of qualitative and quantitative studies on their use.

TYPES OF EVIDENCE

In order to answer the two research questions, all empirical study designs were considered. Academic peer reviewed full-text papers published in printed or electronic format in academic journals or conference proceedings were deemed eligible for inclusion. No language restrictions were applied. Theses, book chapters, non-empirical studies, or unpublished work were excluded.

Data sources and search strategy

Academic databases Scopus, PubMed, and PsycINFO were searched. Additional sources included reference lists of relevant articles and reviews and expert consultation. In the search, strategy terms on "LSB" were combined with terms on dementia. All following terms were being searched in titles, abstracts, and keywords: "life story book" or "life story album" or "storybook" or "life album" or "memory book" or "memory album" or "reminiscence book" or "reminiscence album" or "biography book" or "autobiography book" or "life history book" AND "dementia" or "Alzheimers" or "mci" or "mild cognitive impairment". Two authors (TE and GW) performed the last search run on October 19, 2017.

Study selection

The flow diagram of the search and selection procedure of studies is illustrated in Figure 1. Two data extractors (TE and GW) assessed the eligibility independently in a standardized manner. The 55 retrieved records from the search were screened by title and abstract. An interrater reliability check on the 55 articles was performed, resulting in an agreement of 98% and a Cohen's κ of 0.96, which is considered as "almost perfect" (McHugh, 2012). The disagreement was resolved by consensus, in this case by excluding the article (since no LSBs were used in the intervention). After this first screening, a total of 33 full articles were assessed for eligibility. The extractors read these 33 articles independently. For this second round, the agreement was 91%, with a Cohen's κ of 0.82, which is considered as the upper bound of "strong." The remaining

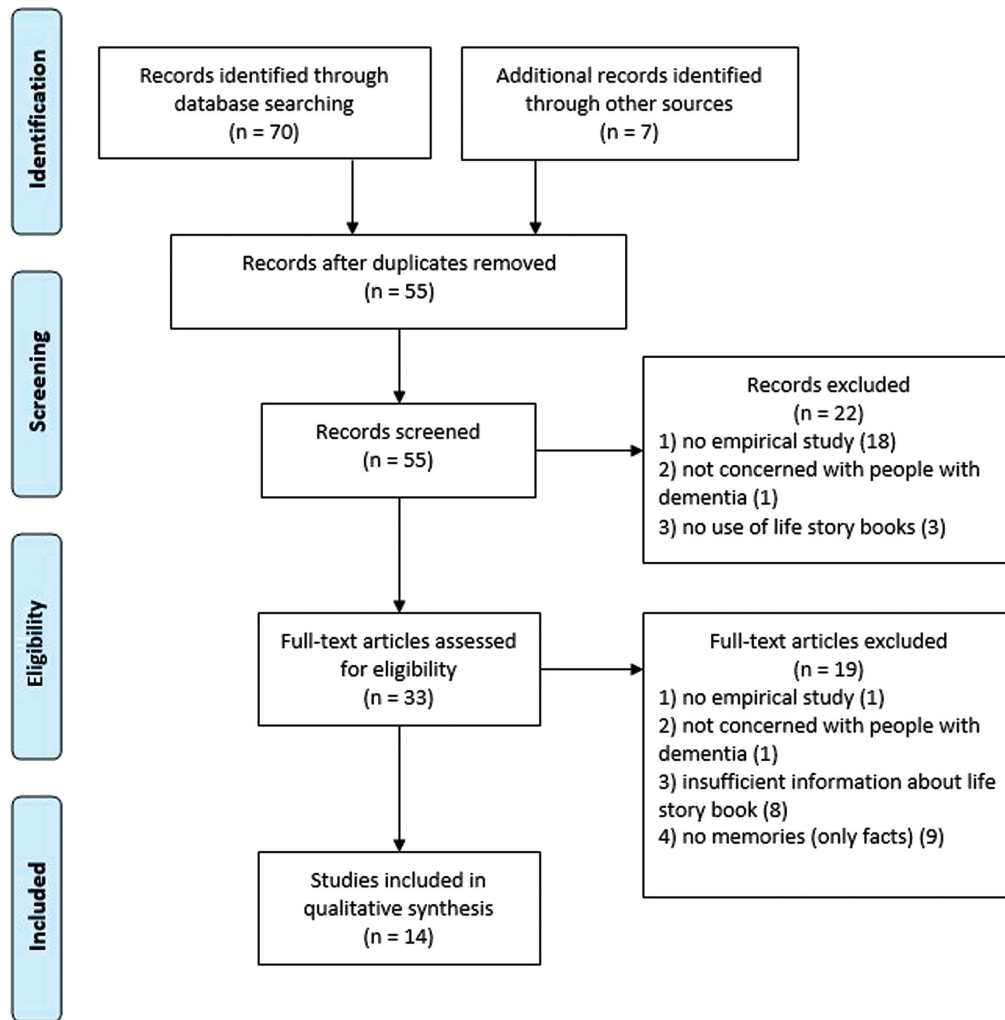


Figure 1. Flow diagram of the search and selection procedure of studies.

three discrepancies were resolved by consensus to exclude the articles (based on the third criterion of insufficient information about the LSB).

Data extraction

In order to answer the first research question *How are LSBs used in dementia care?* the following information was extracted from the articles: target group; type and characteristics of the LSB; implementation of the LSB; and time to create the LSB. To answer the second research question *What are the findings of studies on the use of LSBs?* the following factors were extracted: aim of the study; type of study; sample size; instruments and/or outcome variables; and findings.

Results

Study identification and selection

Figure 1 summarizes the databases hits, exclusion, and final inclusion in a flow diagram. A total of

70 records were found from Scopus (34), PubMed (22), and PsycINFO (14). Reference lists searches and expert consultation added seven studies. After removing duplicates, 55 studies remained and were screened for title and abstract. Based on title and abstract, 22 were discarded as the studies did not meet the inclusion criteria. The full texts of the remaining 33 studies were assessed for eligibility. Of these 33, 19 studies did not meet the inclusion criteria. Finally, a total of 14 studies met the inclusion criteria and were included in the systematic review. Out of the 14 included studies, 12 were identified through database searching.

Study characteristics

All 14 studies were empirical studies published in peer-reviewed journals or conference proceedings. The studies were published between 2003 and 2017. Ten of the 14 studies were conducted in the last five years between 2013 and 2017. In total, 243 persons with dementia participated in the studies.

Research question 1: How are life story books used in dementia care?

To answer this question, first, a general description of the different target groups and type and characteristics of the LSBs are presented in Table 1. Second, Table 2 shows descriptions of the implementation processes and the time needed to create a LSB.

Target group

All included studies concerned people with dementia: Ten studies concerned people with mild to moderate dementia, one study very mild to mild, one mild to severe, one different stages, and one severe dementia. In the study of Crook *et al.* (2016), the participants suffered from mild to moderate dementia and had Down syndrome. A total of nine LSB interventions were carried out for individuals, whereas the other five were dyadic interventions. The mean age varied between 58.6 (Crook *et al.*, 2016) and 92.8 years (Andrews-Salvia *et al.*, 2003). Eleven studies involved both men and women, three only women. The countries in which the studies took place were North Wales (two), United States (solely four; combined with Japan one), United Kingdom (three), Malaysia (two), Korea (one), and Northern Ireland (one). A majority of eight studies was performed in a care home setting, whereas three studies took place at the home situation and two studies in both settings. In one study (Hashim *et al.*, 2013), the setting was not clear.

Type and characteristics of the life story books

In eight studies, a tangible LSB was created. In three studies, both a traditional and another type of LSB was formed, namely a digital book (Ingersoll-Dayton *et al.*, 2016), a rummage box (Crook *et al.*, 2016), and a pen picture (McKeown *et al.*, 2013). In three studies, only a digital version was created: both studies by Hashim *et al.* (2013; 2015) worked with a digital application and the study of Subramaniam and Woods (2016) used previously constructed tangible LSBs as input to create a digital life story movie. A wide variation of materials was used to create the LSBs: photographs, music, narration, stories, blank pages, quotations, and news items. The order of the stories told in the LSBs, when known, was chronological. The length of the books varied from 2 pages (the pen picture) to 70 pages (Subramaniam *et al.*, 2014). The average length of the movies of the Subramaniam and Woods (2016) study was 18 minutes.

Implementation of the life story books

Both the participant and a partner or relative were involved in the process of creating the LSB in five studies. A professional caregiver was additionally helping in five cases. The participant created the book with the professional caregiver – so without a relative – in two cases and in the two remaining cases, it was the other way around: the participant was not involved in the process and the book was created by the relative and a professional caregiver. The researcher helped creating the LSB in the study of Andrews-Salvia *et al.* (2003) and in the control condition of the Subramaniam and Woods study. In five studies, Haight's life review model or life review experience form (LREF; Haight, 1992) was used in the sessions to create the LSB. A total of four studies used the couples life story approach (once combined with techniques from Haight). The number of sessions involved in creating a LSB varied from 3 (Subramaniam and Woods, 2016) to 16 (Subramaniam *et al.*, 2014) with a median of six sessions, while the amount of weeks it took ranged from 9 days (Crook *et al.*, 2016) to 12 weeks (Morgan and Woods, 2010; Subramaniam *et al.*, 2014) and the time per session varied between 15 minutes (Hashim *et al.*, 2015) and 120 minutes (Ingersoll-Dayton *et al.*, 2013). No information was found about actual use, e.g. whether it has been used daily or weekly, for how long, and by whom, of the LSBs after the process of creating it.

Research question 2: What are the designs and findings of studies on the use of life story books?

To answer this question, the main characteristics of the study designs (aims, types of studies, and sample sizes) are presented in Table 3. Table 4 shows the instruments and/or outcome variables used in the studies and presents the findings (qualitative and/or quantitative) of the different studies.

Aim of study

The aims of the studies can be divided into two main categories: (1) getting insight in the challenges and possibilities of implementing LSB interventions in dementia care by evaluating the process and (2) gaining (initial) evidence on the effectiveness of the LSB interventions on participants (e.g. on autobiographical memory and quality of life and mood) and/or their (in)formal caregivers (e.g. on relationship and burden). Seven studies focused on the first aim, three on the second aim, and four had a combination of both aims.

Table 1. Overview of target group and type and characteristics of the life story book.

| AUTHOR | CONDITION OF DEMENTIA | TARGET GROUP | | | | TYPE AND CHARACTERISTICS OF THE LIFE STORY BOOK | | | | |
|---------------------------------------|------------------------------------|----------------------|---------------------|--------|-------------------------|---|---------------------------------------|---|---|----------------------------|
| | | INDIVIDUAL OR DYADIC | MEAN AGE | GENDER | COUNTRY | LIVING SITUATION | TYPE OF LSB | MATERIALS USED | ORDER OF MEMORIES | PAGES/NO OF MEMORIES |
| Kwak <i>et al.</i> (2018) | Very mild to mild | Dyadic | 76.5 | M+F | South Korea | Home and care home | Book | Unknown | Unknown | Unknown |
| Subramaniam and Woods (2016) | Mild to moderate | Individual | 82 | M+F | North Wales | Care home | Movie | Photographs and other visual materials augmented with background music, favorite songs, and narration | Chronological with division in six segments | Average length: 18 minutes |
| Ingersoll-Dayton <i>et al.</i> (2016) | Mild to moderate | Dyadic | 74 (US) 77.4 (J) | M+F | United States and Japan | Home | Traditional (US) and digital book (J) | Photos, stories, and blank pages for the future | Chronological | Unknown |
| Crook <i>et al.</i> (2016) | Mild to moderate and Down syndrome | Individual | 58.6 | M+F | United Kingdom | Care home | Book and rummage box | Photographs and memorabilia | Unknown | Unknown |
| Hashim <i>et al.</i> (2015) | Mild form of AD | Individual | 74 | F | Malaysia | With family | Digital application | Pictures, details, and voice of family members and photographs and descriptions of past events | Unknown | Unknown |
| Subramaniam <i>et al.</i> (2014) | Mild to moderate | Individual | 86 | M+F | North Wales | Care home | Book | Pictures and quotations | Chronological | 50–70 pages |
| Scherrer <i>et al.</i> (2014) | Mild to moderate | Dyadic | 74 | M+F | United States | Home | Book | Photographs, cards, news items, and blank pages for the future | Chronological (assumed) | Unknown |

Table 1. Continued

| AUTHOR | CONDITION OF DEMENTIA | TARGET GROUP | | | | TYPE AND CHARACTERISTICS OF THE LIFE STORY BOOK | | | | |
|---------------------------------------|-----------------------|----------------------|----------|--------|------------------|---|---------------------|---|-------------------------|------------------------|
| | | INDIVIDUAL OR DYADIC | MEAN AGE | GENDER | COUNTRY | LIVING SITUATION | TYPE OF LSB | MATERIALS USED | ORDER OF MEMORIES | PAGES/NO OF MEMORIES |
| Hashim <i>et al.</i> (2013) | Mild form of AD | Individual | 67 | F | Malaysia | Unknown | Digital application | Information and photos of myself, my family, and performing prayer | Unknown | Unknown |
| Ingersoll-Dayton <i>et al.</i> (2013) | Mild to moderate | Dyadic | 75.7 | M+F | United States | Home and care home | Book | Ten photographs or mementoes per chapter (five chapters) | Chronological (assumed) | Max. 50 photos |
| McKeown <i>et al.</i> (2013) | Different stages | Individual | 84.5 | M+F | United Kingdom | Care home | Book or pen picture | Book: photographs and text Pen picture: writing account | Chronological | Pen picture: two pages |
| Morgan and Woods (2010) | Mild to moderate | Individual | 82.5 | M+F | United Kingdom | Care home | Book | Photographs and other materials | Chronological (assumed) | Unknown |
| Haight <i>et al.</i> (2006) | Mild to moderate | Individual | 60–99 | M+F | Northern Ireland | Care home | Book | Photographs and explanatory captions | Chronological (assumed) | Unknown |
| Andrews-Salvia <i>et al.</i> (2003) | Severe | Individual | 92.8 | F | United States | Care home | Book | Division between my life and my family with photographs and declarative sentences | Chronological (assumed) | 20 pages |
| Haight <i>et al.</i> (2003) | No too late | Dyadic | Unknown | M+F | United States | Care home | Book | Pictures, words, and other memorabilia | Unknown | Unknown |

Table 2. Description of the implementation and the time needed to create a life story book

| IMPLEMENTATION OF LIFE STORY BOOK | | | | |
|---------------------------------------|---|--|---|--|
| AUTHOR | THOSE INVOLVED | ROLE OF RESEARCHER(S) | APPROACH | TIME TO CREATE |
| Kwak <i>et al.</i> (2018) | Participant and partner | Engaging participant in process and compiling the book | The couples life story approach which makes use of life review techniques from Haight (1992) | Five weekly sessions |
| Subramaniam and Woods (2016) | Participant and relative | Co-editor of the movie | The life story movie is based on a previously completed conventional book | Creation of movie: on average 8.3 weeks Test prototype: on average three sessions |
| Ingersoll-Dayton <i>et al.</i> (2016) | Participant and partner | Compiling the life story | The couples life story approach | Five weekly sessions |
| Crook <i>et al.</i> (2016) | Participant and family member or other consultee | Facilitating interaction and delivering intervention | Unknown | Nine daily sessions of 30 minutes |
| Hashim <i>et al.</i> (2015) | Participant and caretaker | Explaining and demonstrating the use of the application | Unknown | Eight weekly sessions of 15–30 minutes |
| Subramaniam <i>et al.</i> (2014) | Co-creation: participant and therapist Gift: relative and researcher | Co-creation: therapist as compiler Gift: working close together with relative | Co-creation: based on Haight's life review model and life review experiencing form (LREF; Haight, 1992) | Co-creation: 11–16 sessions in 12 weeks Gift: 5–6 sessions in 12 weeks |
| Scherrer <i>et al.</i> , (2014) | Participant, partner, and social worker | No role in the actual intervention | The couples life story approach with reminiscence sessions with the social worker and the dyad | Five weekly sessions |
| Hashim <i>et al.</i> (2013) | Participant, caretaker, and doctor | Interviewing caretaker and doctor in order to develop content of the application | Unknown | Test prototype: four sessions in four weeks |
| Ingersoll-Dayton <i>et al.</i> (2013) | Participant, partner, and social worker | No role in the actual intervention | The couples life story approach with reminiscence sessions with the social worker and the dyad | Five weekly sessions with an average time span of 76 minutes (40–120) per session |
| McKeown <i>et al.</i> (2013) | Participant, multi-professional staff, and/or family carers | Facilitating the life story work intervention | Life story work | Unknown |
| Morgan and Woods (2010) | Participant and carer or relative | Engaging participant in process and compiling the book | Haight's life review experiencing form (LREF; Haight, 1992) | Approximately 12 weekly sessions of 30–60 minutes |
| Haight <i>et al.</i> (2006) | Participant and care staff (family slightly) | No role in the actual intervention | Haight's life review model and life review experiencing form (LREF; Haight, 1992) | Six weekly sessions of 60 minutes |

Table 2. Continued

| IMPLEMENTATION OF LIFE STORY BOOK | | | | |
|-------------------------------------|--|--|---|--|
| AUTHOR | THOSE INVOLVED | ROLE OF RESEARCHER(S) | APPROACH | TIME TO CREATE |
| Andrews-Salvia <i>et al.</i> (2003) | Family members and researcher | Compiling the book together with family members | Unknown | 12 sessions in three weeks, varying from 7.5 to 22.5 minutes |
| Haight <i>et al.</i> (2003) | Condition 1: both caregiver and care receiver participated separately, but simultaneously Condition 2: only caregiver (without participant) | Condition 1: two reviewers visiting the home Condition 2: unknown | Haight's life review model and life review experiencing form (LREF; Haight, 1992) | Six weekly sessions of 60 minutes |

Hence, there were 11 studies with the first aim and seven with the second aim in total.

Type of study

The 11 studies that evaluated the implementation process used a multiple qualitative case study (three), single case study (two), case-study vignette (three), or another qualitative approach (three). Regarding the seven studies that examined the effectiveness of the LSB interventions, two multiple baseline designs and five (preliminary) (randomized) controlled trial designs were used. Four of these studies compared the LSB intervention to care as usual or no treatment. Of those four, one study compared a LSB with both a no intervention and a rummage box condition (Crook *et al.*, 2016) and another had the following three conditions: (1) life review work carried out separately – but simultaneously – with the person with dementia and the caregiver; (2) life review carried out with the caregiver alone; and (3) an untreated control group (Haight *et al.*, 2003). The fifth controlled trial study compared a co-creation of a LSB with receiving a LSB as a gift (Subramaniam *et al.*, 2014). Participants were randomly assigned to the conditions, except for the Haight *et al.*, 2003 study, for which it remains unclear whether allocation was random.

Sample size

The sample sizes ranged from 1 (the case studies) to 56 (multiple case study; Kwak *et al.*, 2018). For the controlled trials, the average sample size was 20 (with a range from 5 to 31). In four studies, only the persons with dementia were involved in the study, in nine studies informal caregivers, formal caregivers, or other staff were part of the study, and in one study, it was unclear.

Instruments/Outcome variables

A large number of different instruments or outcome variables were used. A distinction can be made between qualitative and quantitative measurements. To explore the implementation process of the interventions, qualitative instruments, such as (semi-structured) interviews, observations, open questions, conversations, and field notes, were used. To assess the effectiveness of the interventions, multiple questionnaires and observational instruments were used on different stakeholders. Participant-focused outcomes were, e.g. autobiographical memory, depression, and quality of life. Furthermore, questionnaires on the quality of the caregiving relationship and communication between the informal caregiver and person with dementia were assessed. Informal caregiver measures were on caregiver burden and formal caregiver measures were on knowledge and attitudes. The outcome measures in the controlled trial studies were applied on baseline as well as during the intervention (Crook *et al.*, 2016) or after the intervention with a maximum of 18 weeks (Subramaniam *et al.*, 2014).

Findings

QUALITATIVE FINDINGS

All those involved looked back on the interventions as an enjoyable process and they viewed a LSB as a useful tool triggering memories and – largely positive – emotions. Participants, relatives, and care staff saw the value of the LSB mainly in improvements in relationships: partner affirmation, engagement, fullness of life as a couple, social interaction, and communication. Furthermore, several (cultural) themes were identified and recommendations for implementing a LSB intervention were given, e.g.

Table 3. Overview of study characteristics

| AUTHOR | AIM OF STUDY | TYPE OF STUDY | SAMPLE SIZE |
|---------------------------------------|--|---|----------------|
| Kwak <i>et al.</i> (2018) | Adaptation of “couples life story approach” in South Korea | Multiple qualitative case study design | 56 |
| Subramaniam and Woods (2016) | Acceptability and efficacy | Multiple baseline design | 6 |
| Ingersoll-Dayton <i>et al.</i> (2016) | Development of “couples life story approach” | Qualitative study | 29 |
| Crook <i>et al.</i> (2016) | Initial evidence on well-/ill-being and behavior | Qualitative study | 5 |
| Hashim <i>et al.</i> (2015) | Randomized multiple baseline design with three conditions: 1) life story book 2) rummage box 3) no-intervention condition | Qualitative case study | 1 |
| Subramaniam <i>et al.</i> (2014) | Evidence on management of everyday tasks, reminiscence, and cognitive function Effect of different pathways for developing a life story book | Preliminary RCT with two conditions: 1) receiving 12 individual life review sessions and co-creating a LSB 2) receiving a personal LSB created by their relatives as a “gift” | 23 11 12 |
| Scherrer <i>et al.</i> (2014) | Mapping challenges of conducting narrative-based interventions | Two case study vignettes | 20 |
| Hashim <i>et al.</i> (2013) | Developing a digital memory book application to the need of the patient | Multiple baseline single case design | 1 |
| Ingersoll-Dayton <i>et al.</i> (2013) | To help individuals who have dementia and their spouses or partners communicate and reminisce about their life and develop a book that incorporates mementoes of their life | Qualitative case study | 24 |
| McKeown <i>et al.</i> (2013) | To understand experiences of people with dementia, family carers, and care staff in using life story work and to explore the process taken for life story work to be implemented | Qualitative study | 4 |
| Morgan and Woods (2010) | To gain empirical evidence on the impact of life review therapy with people with dementia | Multiple baseline design | 17 8 9 |
| Haight <i>et al.</i> (2006) | To test the effectiveness of a structured life review/life storybook process | Preliminary RCT with two conditions: 1) life review which culminated in the creation of a LSB 2) treatment as usual Two case study vignettes Controlled pilot RCT with two conditions: 1) life review/life storybook 2) care as usual | 31 15 16 |

Table 3. Continued

| AUTHOR | AIM OF STUDY | TYPE OF STUDY | SAMPLE SIZE |
|-------------------------------------|--|---|-------------------|
| Andrews-Salvia <i>et al.</i> (2003) | To assess the effect of memory books on the number of on-topic facts stated for three topics (life, family, and day) | Multiple baseline design | 4 |
| Haight <i>et al.</i> (2003) | Reviewing the application of reminiscence with people with dementia | Study with three conditions: 1) life review work carried out separately – but simultaneously – with the person with dementia and the caregiver 2) life review carried out with the caregiver alone 3) an untreated control group Two case study vignettes | 22 - - - |

Note: - = missing data.

on how to incorporate difficult life events in the LSB, how to tell a mutual story, and how to end the story. No negative effects were reported.

QUANTITATIVE EFFECTS

In the five controlled trial studies, the LSB interventions showed significant improvements in autobiographical memory, mood, depression, and quality of life of the persons with dementia compared with care as usual or no treatment. Furthermore, the communication and quality of relationships between participants and their informal caregivers improved significantly. Finally, significant improvement on staff attitudes and knowledge was found. In one study, two reminiscence intervention conditions (one being a LSB intervention and the other a rummage box) showed significant improvement compared to the no intervention condition, but no difference was found between the LSB condition and the rummage box condition (Crook *et al.*, 2016). Moreover, some positive non-significant changes in independence and behavior problems in favor of the LSB intervention were reported (Haight *et al.*, 2006). One study reported a mixed outcome: the persons with dementia in the dyadic LSB condition showed a significant decrease in cognition compared to the caregiver only and no treatment condition, while mood and burden improved in both the dyadic and caregiver only condition (Haight *et al.*, 2003).

Discussion

Reminiscence can be used to enhance PCC and has proven to be effective for persons with dementia (Woods *et al.*, 2005; Subramaniam and Woods, 2012; Blake, 2013; Huang *et al.*, 2015).

The creation of a LSB is a specific form of reminiscence that is the result of a life review process that illustrates the biography of a person. This systematic review of 14 studies showed that most LSBs were tangible books that were created in about six individual sessions in nursing homes with persons with varying degrees of dementia as well as with their informal and formal caregivers. Process evaluations showed the value of LSBs in triggering memories and positive emotions and in improving the relation with the person with dementia. Quantitative evaluations supported this value as significant improvements were found on autobiographical memory, depression, mood, and quality of life of the persons with dementia, as well as on the quality of relationships and communication between the person with dementia and the informal caregiver. Furthermore, effects on burden of the informal caregivers and on attitudes and knowledge of formal caregivers were found in comparison to care as usual.

The findings for persons with dementia are in line with earlier reviews and a meta-analysis that have shown that reminiscence activities can contribute to cognitive functioning, depression, and quality of life of persons with dementia (Woods *et al.*, 2005; Subramaniam and Woods, 2012; Blake, 2013; Huang *et al.*, 2015). The findings for caregivers and their relation to persons with dementia indicate that LSBs can contribute to the culture change towards more PCC (Koren, 2010; AGS, 2016; Higgs and Gilleard, 2017). The relationship between persons with dementia and their formal and informal caregivers appears to be improved and the expression of values and preferences of persons with dementia in LSBs can further contribute to support their health and life goals. This also aligns with previous findings that

Table 4. Overview of instruments and findings

| AUTHOR | INSTRUMENTS/OUTCOME VARIABLES | FINDINGS |
|---------------------------------------|---|--|
| Kwak <i>et al.</i> (2018) | Primary data: session notes written by interventionists. Supplementary data: interviews with participants and weekly team meeting notes | Cultural themes identified: (1) dealing with negative memories in early years of marriage; (2) communication styles and patterns; (3) ways to incorporate difficult life events into the life story book; and (4) complex dynamics of hierarchy in the relationship between older couples and the interventionist |
| Subramaniam and Woods (2016) | Quantitative: Quality of life-Alzheimer's disease scale (QOL-AD); autobiographical memory interview extended version (AMI-E); subscales PSS and AIS); Geriatric Depression Scale Residential (GDS-12R); quality of the caregiving relationship questionnaire (QCPR). | Digital LSB > LSB. Positive average improvement on quality of life, autobiographical memory (subscale PSS), depression, and quality of caregiving relationship |
| Ingersoll-Dayton <i>et al.</i> (2016) | Qualitative: Open-ended questions. Clinical analysis of the progress of the couples discussed by members of the Japanese and American teams. Based on these discussions, four themes emerged that characterized how the couples experienced this intervention | Those involved viewed digital life story books as a very useful tool stimulating memories, triggering positive emotions, and encouraging conversation and interaction Themes found: partner affirmation (highlighting each other's strengths), improved engagement; handling losses; and fullness of a life as a couple |
| Crook <i>et al.</i> (2016) | Dementia care mapping (DCM), divided into behavior category code (BCC), and mood-engagement value (ME), calculated into well-/ill-being (WIB). The questionnaires were filled out on two days before baseline and on nine consecutive days during the intervention conditions | Reminiscence conditions > no intervention condition. Higher WIB, but not consistent across all participants. Both reminiscence conditions tended to be associated with an increase in communicative, expressive, and intellectual behaviors. No significant difference was found between the LSB and rummage box |
| Hashim <i>et al.</i> (2015) | An evaluation form that contains a set of questions related to presentation, motivation, understanding, memorability, learnability, and usability | Positive feedback and user satisfaction. Patient felt motivated and enjoyed using the application that supported managing her daily activities, reminiscence, and cognitive function |
| Subramaniam <i>et al.</i> (2014) | Quality of life-Alzheimer's disease (QOL-AD); autobiographical memory interview extended version (AMI-E); The Geriatric Depression Scale (Residential) (GDS-12R); quality of the caregiving relationship questionnaire (QCPR participant and relative); approaches to dementia questionnaire (ADQ); staff knowledge of care-recipient questionnaire. The questionnaires were filled out on baseline and on follow-up at 12 and 18 weeks Description of two cases | Both LSB conditions (co-creation and gift) show significant improvements on quality of life ($p = 0.035$) and autobiographical memory ($p = 0.001-0.005$) at post-intervention, and on quality of relationship at six-week follow-up ($p = 0.046$). Staff attitude ($p < 0.001$) and knowledge ($p < 0.001$) was improved at six-week follow-up. No difference was found between the two LSB conditions The book appeared to play a role as a maintenance tool after the completion of the life review process and helped the participants to look back on their life |
| Scherrer <i>et al.</i> (2014) | During team meetings, social workers described the positive changes that they observed in their weekly meetings with couples, as well as the challenges they encountered. These challenges were discussed as the project leaders and other members of the project team considered strategies to address them | Recommendations are given on how to (1) construct a narrative from disparate stories, (2) tell a mutual story, (3) tell the story of a couple that has been in a shorter relationship, (4) incorporate others in the story, (5) include difficult life moments, and (6) end the story |

Table 4. Continued

| AUTHOR | INSTRUMENTS/OUTCOME VARIABLES | FINDINGS |
|---------------------------------------|--|--|
| Hashim <i>et al.</i> (2013) | Interviews with caretaker and doctor to assess needs; observation of behavior during sessions; testing of long- and short-term memory (unclear how) | The results show that by using the application, not only the patient's reminiscence is improved regarding to performing prayer (short term memory), but it also upgrades the social interaction and communication between the patient and caretaker |
| Ingersoll-Dayton <i>et al.</i> (2013) | Questionnaires with open-ended questions about their reactions to the approach (participants and caregivers) and observations | Positive aspects mentioned by participants are as follows: enjoyed reliving story of life together; communication tips were useful; enjoyed the life story book; planned to share the life story book with others; meaningful engagement; and helped memory |
| McKeown <i>et al.</i> (2013) | Semi-structured interviews, observation, conversations, and field notes | Private memories were sometimes recalled by the person with dementia that were not for inclusion in any written product; enabling the person with dementia to tell their own life story could be a challenge; quality of the life story books was variable; and, at times, life story work may be overused with the person with dementia |
| Morgan and Woods (2010) | Geriatric Depression Scale–Short Form (GDS–SF); autobiographical memory interview (AMI). The questionnaires were filled out on baseline and on follow-up at six weeks | LSB>treatment as usual. Significant improvement on depression (p=0.009) and autobiographical memory (p=0.016) during follow-up |
| Haight <i>et al.</i> (2006) | Mini Mental Status Exam (MMSE); Cornell Scale for Depression (CSDD); Alzheimer's Mood Scale N & P (neg. and pos.); functional independence measure (FIM); Communication Observation Scale for Cognitively Impaired (CS); memory and behavior problems checklist (MBS). The questionnaires were filled out one week before and one week after the intervention (of six weeks) | Although the life review process and creation of the LSB was difficult, the participants enjoyed it |
| Andrews-Salvia <i>et al.</i> (2003) | Number of on-topic facts made by the subjects during conversation with the experimenters using the memory books as memory aids | LSB > care as usual. Significant improvement on cognitive functioning (p < 0.0005), depression (p < 0.015), positive mood (p < 0.008), and communication (p < 0.005). Improvement (not significant) on independence and memory and behavior problems |
| Haight <i>et al.</i> (2003) | Mini-Mental State Exam (MMSE); Alzheimer Mood Scale (extracted from transcribed interviews); revised memory and behavior problem checklist; burden interview. The questionnaires were filled out on baseline and on follow-up at two months | All subjects stated more on-topic facts using the memory books than during the baseline condition |
| | Descriptions of two cases | Dyadic < caregiver only & no treatment. Significant decrease on cognition (p < 0.03). Dyadic > caregiver only & no treatment. Significant improvement on mood (p < 0.04). Dyadic & caregiver only > no treatment. Significant increased burden (p < 0.06) and behavior problems (p < 0.05) in no treatment The case studies helped to elucidate the process |

remembrance has the potential to enable care staff to see the person behind the patient and enable the patient's voice to be heard, verbally and non-verbally (Woods *et al.*, 2005).

According to the framework for trials of complex interventions created by the Medical Research Council (MRC, 2000), the research on LSBs is ranged between the phase of modelling and the phase of exploratory trials (phase I and phase II). The use of LSBs and the qualitative studies provides insight in the components and underlying mechanisms of LSBs (phase I). The main mechanism is the recollection and sharing of autobiographical memories. The components are an individual or dyadic life review that results in a tangible (digital) life story. The quantitative studies in this review apply to the exploratory trial phase (phase II). In this phase, the components are tested in different designs. Small sample sizes and non-random allocation of participants are characteristic for this phase. Despite the fact that the majority of the controlled trials in this review were randomized, we need to interpret the significant improvements found on several outcomes in this review with caution, especially due to the small sample sizes.

The next phase (phase III) would be to conduct larger randomized controlled trial (RCT) studies to establish the effects of LSBs on people with dementia. This phase asks for a good choice of intervention characteristics in relation to outcome measures. Given the diversity in aims and methods of the included interventions, it is hard to state one as the best practice at this moment in time. Hence, when conducting a larger trial, a clear formulation of the aims of the LSB intervention and how the specific components contribute to the aims is necessary.

Overall, the research in the field of the use of LSBs in dementia care is in the first phases of providing evidence. However, one has to realize that research on LSBs in dementia care is a fairly new area of interest, hence the lack of good, solid RCTs with large sample sizes. Given the current state, the small RCTs, pilot studies, and qualitative case studies of this review do provide insights and help future research.

This review shows that an intervention may focus more on the person with dementia and try to improve autobiographical memory, mood, depression, and/or quality of life or, alternatively, focus more on the relationships of (in)formal caregivers with the person with dementia. The process of creating a LSB may be different for both purposes, for example, in terms of the persons who are involved in the process, or in the use of individual forms like Haight LREF (Haight, 1992) versus a dyadic approach like the couples

life story approach (Ingersoll-Dayton *et al.*, 2013). Technology might contribute to the first aim due to multimedia effects like music or movies that may more strongly involve the person with dementia in the process of recollecting memories and improving mood. Technology might contribute in another way to the second aim as different persons can contribute to the creation and use of digital LSBs more easily, also making the LSB more interactive. Is it only after conducting good trials that the last phase (phase IV) of long-term implementation can be realized?

This review is the first to provide an overview on the use of LSBs as a specific domain within the field of reminiscence in dementia care. The studies included were diverse in their aims and consequently in their methods, which makes it difficult to compare the studies in a consistent way. However, the diversity does show that creating LSBs needs to be a tailored process. Especially in a time when the need for more PCC is greater than ever, one could question whether there is one golden standard since it always will be a personal, individualized process to create one's life story. Nevertheless, it is worthwhile to investigate the effect of personalized LSBs on larger studies.

Since 12 of the 14 included studies were identified through database searching, it is assumed that the constructed search strategy was sufficient and all relevant studies were identified. The majority of studies in this review report on positive findings or effects, although some studies do mention less positive outcomes. Failed attempts on studies on LSBs may not have been published. This systematic review shows indications of effects of creating LSBs and possible ways to implement LSBs in dementia care, but information on their actual use after the creating process and long-term effects after implementation is lacking.

This systematic review shows that research on LSB interventions for people with dementia is emerging and confirms that the use of LSBs to support reminiscence and PCC is promising.

Conflict of interest

None.

Description of authors' roles

T. Elfrink and G. Westerhof formulated the research questions and were the data extractors of this review. T. Elfrink analyzed the data and wrote the article with supervision and feedback from G. Westerhof, M. Kunz, and S. Zuidema. All authors provided comments and read and approved the final manuscript.

Acknowledgments

This research was supported by a grant from ZonMw, Alzheimer Nederland, & PGGM. Contract grant number: 733050610.

References

- American Geriatrics Society Expert Panel on Person-Centered Care (AGS).** (2016). Person-centered care: a definition and essential elements. *Journal of the American Geriatrics Society*, 64, 15–18. doi: 10.1111/jgs.13866.
- Andrews-Salvia, M., Roy, N. and Cameron, R. M.** (2003). Evaluating the effects of memory books. *Journal of Medical, Speech-Language Pathology*, 11, 51–59.
- Blake, M.** (2013). Group reminiscence therapy for adults with dementia: a review. *British Journal of Community Nursing*, 18, 228–233. doi: 10.12968/bjcn.2013.18.5.228.
- Clarke, A.** (2000). Using biography to enhance the nursing care of older people. *British Journal of Nursing*, 9, 429–433. doi: 10.12968/bjon.2000.9.7.6323.
- Clarke, A., Jane Hanson, E. and Ross, H.** (2003). Seeing the person behind the patient: enhancing the care of older people using a biographical approach. *Journal of Clinical Nursing*, 12, 697–706. doi: 10.1046/j.1365-2702.2003.00784.
- Conde-Sala, J. L. et al.** (2016). Effects of anosognosia and neuropsychiatric symptoms on the quality of life of patients with Alzheimer's disease: a 24-month follow-up study. *International Journal of Geriatric Psychiatry*, 31, 109–119. doi: 10.1002/gps.4298.
- Crook, N., Adams, M., Shorten, N. and Langdon, P. E.** (2016). Does the well-being of individuals with Down syndrome and dementia improve when using life story books and rummage boxes? a randomized single case series experiment. *Journal of Applied Research in Intellectual Disabilities*, 29, 1–10. doi: 10.1111/jar.12151.
- De Vugt, M. E. et al.** (2003). Behavioural disturbances in dementia patients and quality of the marital relationship. *International Journal of Geriatric Psychiatry*, 18, 149–154. doi: 10.1002/gps.807.
- Haight, B. K.** (1992). The structured life-review process: a community approach to the ageing client. In G. M. M. Jones and B. M. L. Miesen (eds.), *Care-Giving in Dementia* (pp. 272–292). London: Routledge.
- Haight, B. K., Bachman, D. L., Hendrix, S., Wagner, M. T., Meeks, A. and Johnson, J.** (2003). Life review: treating the dyadic family unit with dementia. *Clinical Psychology & Psychotherapy*, 10, 165–174. doi: 10.1002/cpp.367.
- Haight, B. K., Gibson, F., and Michel, Y.** (2006). The Northern Ireland life review/life storybook project for people with dementia. *Alzheimer's & Dementia*, 2, 56–58. doi: 10.1016/j.jalz.2005.12.003.
- Hashim, A. H. A., Ismail, A. N., Rias, R. M. and Mohamed, A.** (2015). The development of an individualized digital memory book for Alzheimer's disease patient: a case study. In *International Symposium Technology Management and Emerging Technologies (ISTMET), 2015* (pp. 227–232). Langkawi Island, Malaysia: IEEE. doi: 10.1109/ISTMET.2015.7359034.
- Hashim, A. H. A., Rias, R. M. and Kamaruzaman, M. F.** (2013). The use of personalized digital memory book as a reminiscence therapy for Alzheimer's disease (AD) patients. In H. B. Zaman, P. Robinson, P. Olivier, T. K. Shih, and S. Velastin (eds.), *International Visual Informatics Conference* (pp. 508–515). New York: Springer-Verlag. doi: 10.1007/978-3-319-02958-0_46.
- Higgs, P. and Gilleard, C.** (2017). Ageing, dementia and the social mind: past, present. *Sociology of Health & Illness*, 39, 175–181. doi: 10.1111/1467-9566.12536.
- Huang, H. C. et al.** (2015). Reminiscence therapy improves cognitive functions and reduces depressive symptoms in elderly people with dementia: a meta-analysis of randomized controlled trials. *Journal of the American Medical Directors Association*, 16, 1087–1094. doi: 10.1016/j.jamda.2015.07.010.
- Ingersoll-Dayton, B., Spencer, B., Campbell, R., Kurokawa, Y. and Ito, M.** (2016). Creating a duet: the couples life story approach in the United States and Japan. *Dementia*, 15, 481–493. doi: 10.1177/1471301214526726.
- Ingersoll-Dayton, B., Spencer, B., Kwak, M., Scherrer, K., Allen, R. S. and Campbell, R.** (2013). The couples life story approach: a dyadic intervention for dementia. *Journal of Gerontological Social Work*, 56, 237–254. doi: 10.1080/01634372.2012.758214.
- Koren, M. J.** (2010). Person-centered care for nursing home residents: the culture-change movement. *Health Affairs*, 29, 312–317. doi: 10.1377/hlthaff.2009.0966.
- Kwak, M., Ha, J. H., Hwang, S. Y., Ingersoll-Dayton, B. and Spencer, B.** (2018). Cultural adaptation of a dyadic intervention for Korean couples coping with Alzheimer's disease. *Clinical Gerontology*, 41, 217–226. doi: 10.1080/07317115.2017.1366385.
- McHugh, M. L.** (2012). Interrater reliability: the kappa statistic. *Biochemia Medica*, 22, 276–282. doi: 10.11613/BM.2012.031.
- McKeith, I. and Cummings, J.** (2005). Behavioural changes and psychological symptoms in dementia disorders. *The Lancet Neurology*, 4, 735–742. doi: 10.1016/S1474-4422(05)70219-2.
- McKeown, J., Clarke, A. and Repper, J.** (2006). Life story work in health and social care: systematic literature review. *Journal of Advanced Nursing*, 55, 237–247. doi: 10.1111/j.1365-2648.2006.03897.x.
- McKeown, J., Ryan, T., Ingleton, C. and Clarke, A.** (2013). 'You have to be mindful of whose story it is': the challenges of undertaking life story work with people with dementia and their family carers. *Dementia*, 14, 238–256. doi: 10.1177/1471301213495864.
- Medical Research Council.** (2000). *A Framework for the Development and Evaluation of RCTs for Complex Interventions to Improve Health*. London: MRC.
- Moher, D. et al.** (2015). Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015 statement. *Systematic Reviews*, 4, 1. doi: 10.1186/2046-4053-4-1.
- Morgan, S. and Woods, R. T.** (2010). Life review with people with dementia in care homes: a preliminary

- randomized controlled trial. *Non-pharmacological Therapies in Dementia*, 1, 43–60.
- Peeters, J. E., Werkman, W. and Francke, A. L.** (2012). *Dementiemonitor Mantelzorg: Problemen, Zorgbehoeften, Zorggebruik en Oordelen van Mantelzorgers*. Utrecht, the Netherlands: Nivel.
- Prince, M., Prina, M. and Guerchet, M.** (2013). *Journey of Caring. An Analysis of Long-Term Care for Dementia*. London: Alzheimer's Disease International.
- Prince, M., Wimo, A., Guerchet, M., Ali, G. C., Wu, Y. T. and Prina, A.M.** (2015). *The Global Impact of Dementia. An Analysis of Prevalence, Incidence, Cost and Trends*. London: Alzheimer's Disease International.
- Scherrer, K. S., Ingersoll-Dayton, B. and Spencer, B.** (2014). Constructing couples' stories: narrative practice insights from a dyadic dementia intervention. *Clinical Social Work Journal*, 42, 90–100.
- Subramaniam, P. and Woods, B.** (2012). The impact of individual reminiscence therapy for people with dementia: systematic review. *Expert Review of Neurotherapeutics*, 12, 545–555. doi: 10.1586/ern.12.35.
- Subramaniam, P. and Woods, B.** (2016). Digital life storybooks for people with dementia living in care homes: an evaluation. *Clinical Interventions in Aging*, 11, 1263–1276. doi: 10.2147/CIA.S111097.
- Subramaniam, P., Woods, B. and Whitaker, C.** (2014). Life review and life story books for people with mild to moderate dementia: a randomised controlled trial. *Aging & Mental Health*, 18, 363–375. doi: 10.1080/13607863.2013.837144.
- VandenBos, G. R.** (eds.) (2006). *APA Dictionary of Psychology*, 1st edn. Washington, DC: American Psychological Association.
- Woods, B., Spector, A., Jones, C., Orrell, M. and Davies, S.** (2005). Reminiscence therapy for dementia. *Cochrane Database of Systematic Reviews*, 2. doi: 10.1002/14651858.CD001120.pub2.
- World Health Organization (WHO)**. (2017). *Facts on Dementia*. Available at: <http://www.who.int/features/factfiles/dementia/en/>; last accessed 9 May 2017.

REVIEW

Neurobiological findings associated with high cognitive performance in older adults: a systematic review

Wyllians Vendramini Borelli, Lucas Porcello Schilling, Graciane Radaelli, Luciana Borges Ferreira, Leonardo Pisani, Mirna Wetters Portuguez and Jaderson Costa da Costa

Brain Institute of Rio Grande do Sul (BraIns), Pontifical Catholic University of Rio Grande do Sul, Porto Alegre, Brazil

ABSTRACT

Objectives: to perform a comprehensive literature review of studies on older adults with exceptional cognitive performance.

Design: We performed a systematic review using two major databases (MEDLINE and Web of Science) from January 2002 to November 2017.

Results: Quantitative analysis included nine of 4,457 studies and revealed that high-performing older adults have global preservation of the cortex, especially the anterior cingulate region, and hippocampal volumes larger than normal agers. Histological analysis of this group also exhibited decreased amyloid burden and neurofibrillary tangles compared to cognitively normal older controls. High performers that maintained memory ability after three years showed reduced amyloid positron emission tomography at baseline compared with high performers that declined. A single study on blood plasma found a set of 12 metabolites predicting memory maintenance of this group.

Conclusion: Structural and molecular brain preservation of older adults with high cognitive performance may be associated with brain maintenance. The operationalized definition of high-performing older adults must be carefully addressed using appropriate age cut-off and cognitive evaluation, including memory and non-memory tests. Further studies with a longitudinal approach that include a younger control group are essential.

Key words: memory, aging, magnetic resonance imaging

Abbreviations

| | |
|------|------------------------------|
| PET | Positron Emission Tomography |
| PIB | Pittsburgh compound B |
| DVR | Distribution volume ratio |
| AD | Alzheimer's disease |
| ApoE | Apolipoprotein E |

Introduction:

The incidence of dementia has increased in direct proportion to aging in the general population leading to a massive worldwide impact (Prince *et al.*, 2015). As 99.6% of drug therapies for Alzheimer's

disease (AD) have not provided promising results (Cummings *et al.*, 2014), different therapeutic targets must be investigated. On the extreme opposite of the cognitive continuum, "Superaging" has become a rising subject of interest as some older adults show exceptional memory ability (Rogalski *et al.*, 2013). Accordingly, individuals that achieve a successful cognitive aging trajectory can either experience less pathological alterations in their brains or show resistance to age-related physiological decline. These older adults with high cognitive performance may exhibit structural and molecular mechanisms that ultimately lead to unusually preserved brain functioning throughout the lifespan.

Older adults tend to show an increased variability of cognitive functions during the aging process (Hedden and Gabrieli, 2004). Currently, many theories of successful aging attempt to explain this vast cognitive variability in older age. There

Correspondence should be addressed to: Jaderson Costa da Costa, MD, Ph.D., Brain Institute of Rio Grande do Sul (BraIns), Pontifical Catholic University of Rio Grande do Sul, Av. Ipiranga, 6690, Porto Alegre, RS, 90610-000, Brazil. Phone: 5551 3320 5959. Email: jcc@puers.br. Received 20 Nov 2017; revision requested 3 Jan 2018; revised version received 16 Feb 2018; accepted 5 Mar 2018. First published online 18 April 2018.

are two main theories regarding healthy cognitive aging: the reserve concepts (Stern, 2009) and the brain maintenance (Nyberg *et al.*, 2012). The concept of cognitive and brain reserves has been put forward to explain differences in cognitive decline among older adults, supposed to be a consequence of increased neuronal count and size (Stern, 2009). The amount of reserve may determine the impact of pathological age-related alterations on cognitive and structural phenotypes. However, this definition does not explain why some older adults show cognitive and brain preservation through aging (Habeck *et al.*, 2016).

As a complementary hypothesis to the notion of reserve, Nyberg *et al.* (2012) introduced the notion of brain maintenance. In this conception, structural and functional brain maintenance determines the preservation of memory and other cognitive functions across the lifespan. It poses the avoidance or minimization of the aging brain alterations as best predictors of successful memory abilities in late-life. However, few studies have focused on the biological basis of brain maintenance and its consequences on cognitive aging. Herein, we aim to perform a systematic literature review of studies with older adults with superior cognitive ability to investigate neurobiological findings associated with successful cognitive aging.

Methods

This review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Statement (Moher *et al.*, 2009) and was registered at the International Register of Prospective Systematic Reviews, under identification number 42017053255.

Eligibility criteria

LITERATURE SEARCH

We performed a search in MEDLINE and Web of Science for pertinent data from January 2002 to November 2017. As we aimed to provide an overview of all available literature, peer-reviewed journals, and grey literature were investigated.

The search strategy included the following key terms: “successful cognitive aging,” “high-performing older adults,” “SuperAgers,” and “exceptional memory capacity.” Search terms in Medline also included any of the following Medical Subject Headings (MeSH), and term combinations indicated by “AND” and “OR” were used as Boolean operators: successful OR exceptional OR excellent OR high-performing AND cognition OR cognitive OR memory OR

brain AND aging OR superaging OR older adults OR elders OR superagers OR supernormals. The Boolean operators were not used in the Web of Science search due to the structure of its search engine. There were no language restrictions. A meta-analysis was not deemed possible in the present work because of the heterogeneity of the data and the limited number of studies.

STUDY SELECTION

Two authors (LBF and LP) independently assessed potentially eligible studies for their suitability for inclusion in the review. We resolved any disagreements by discussion or by a third reviewer (WVB). During the screening of titles and abstracts, relevant papers were defined if they mentioned aspects of high cognitive ability, such as “exceptional memory,” “exceptional cognition,” “excellent memory,” and “high-performing.” Abstracts were analyzed according to the inclusion criteria, and all studies that met these criteria were included for full article reading.

To recognize subjects within the top level of cognitive capacity in older age, the inclusion criteria were rigorously determined. Articles were required to (1) show original data, (2) include a group of adults who were 70 years of age or older, (3) clearly describe the inclusion criteria for participants, and (4) include individuals in the high-performing group with cognitive score higher than age-matched peers or than that expected for their age group based on normative data. Exclusion criteria were as follows: (1) No clinical characteristics were available, (2) no standardized neuropsychological criteria were used, and (3) any qualitative study.

EXTRACTION OF DATA

Data extraction was conducted by two authors (LP and LBF) from papers that met the inclusion criteria and included the following: demographic characteristics of the sample, the definition used for classifying the high-performing older group, neuropsychological assessments, other inclusion criteria, and main outcomes of each study. To better suit the proposed review, we included only studies with standardized neuropsychological assessment.

Results

Characteristics of included articles

From 4,457 potentially relevant citations retrieved from electronic databases and searches of reference lists, nine (0.2%) studies met the inclusion criteria (Figure 1). There were three studies on

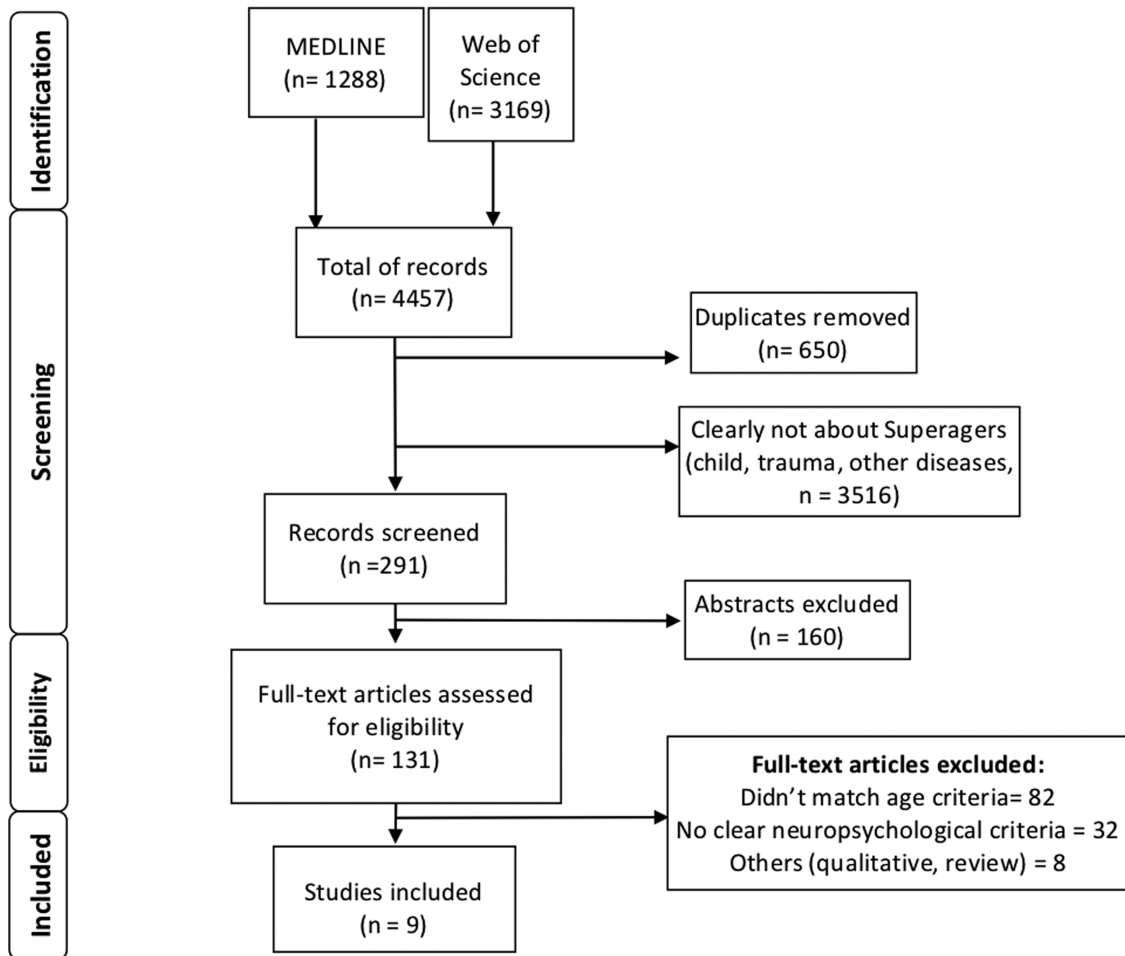


Figure 1. Flow chart of the review.

neuroimaging (Harrison *et al.*, 2012; Cook *et al.*, 2017; Dekhtyar *et al.*, 2017), two on histological analysis (Gefen *et al.*, 2015; Janeczek *et al.*, 2017), one on plasma metabolites (Mapstone *et al.*, 2017), and two on neuropsychological profile (Gefen *et al.*, 2014; Cook Maher *et al.*, 2017). One study reported findings that had been previously published, provided another specific outcome, namely apolipoprotein E (ApoE) status (Rogalski *et al.*, 2013) (Table 1). Sun *et al.* (Sun *et al.*, 2016) cited the term “SuperAgers” but did not match the age criteria.

Studies that met the eligibility criteria provided a neuropsychological profile of high-performing older adults using either validated tests or at least one control group (Table 2). Sample sizes were related to the type of study (range: 5–330) and all studies reported clinical, neurological, and/or psychiatric screening criteria to confirm a healthy sample. Imaging studies were controlled for sex, age, and education, except Harrison (Harrison *et al.*, 2012) that does not mention the gender of included individuals. Mapstone *et al.* (2017)

used a composite Z-score adjusted for sex, age, and education. Histologic outcomes (Gefen *et al.*, 2015; Janeczek *et al.*, 2017) were analyzed only in high-performing females, while the control group included both genders, and Rogalski *et al.* (2013) did not mention this information for ApoE analysis. As seven of the nine studies were conducted by researchers from Northwestern University, the total sample included in this review may overlap some individuals. There were a total of 199 individuals with collected data.

Notably, high-performing older adults were described with different terms, namely “SuperAgers” (Harrison *et al.*, 2012; Rogalski *et al.*, 2013; Gefen *et al.*, 2014; 2015; Cook *et al.*, 2017; Cook Maher *et al.*, 2017; Janeczek *et al.*, 2017), “Supernormals” (Mapstone *et al.*, 2017), and “Optimal performers” (Dekhtyar *et al.*, 2017). All definitions converged in classifying older adults according to their episodic memory performance. The Rey Auditory-Verbal Learning Test was employed in eight of the nine studies, and one study used a composite memory score that included the Memory Capacity Test

Table 1. Summary of included articles

| CITATION | TYPE OF OUTCOME | CHARACTERISTICS OF PARTICIPANTS (N) | DEFINITION OF THE HIGH-PERFORMING OLDER GROUP | OTHER INCLUSION CRITERIA | TESTS PERFORMED | MAIN OUTCOMES |
|-------------------------------|-----------------|--|---|---|------------------------|--|
| Harrison <i>et al.</i> (2012) | Structural MRI | HP (12): mean age = 83.5 (3), mean years of education = 14.8 (2.4). YG (14): mean age = 57.9 (4.3), mean years of education = 16.1 (2.9). NC (10): mean age = 83.1 (3.4), mean years of education = 17.5 (2.2) | Age \geq 80 years Perform at or above average normative values for individuals in their 50s and 60s (RAVLT) delayed-recall raw score \geq 9) and within one standard deviation of the average for the non-memory measures | To have preserved activities of daily living and lacked clinical or structural evidence of neurologic or psychiatric disease | RAVLT; BNT; TMT-B; CFT | HP=YG>NC in whole brain volume HP>YG>NC in left anterior cingulate volume |
| Rogalski <i>et al.</i> (2013) | ApoE pattern | HP (12): mean age = 83.5 (3). NC (330): median age = 70 | Age \geq 80 years Perform at or above average normative values for 50–60 yo (RAVLT) delayed-recall raw score \geq 9) and within one standard deviation of the average for the non-memory measures | To have preserved activities of daily living and lacked clinical or structural evidence of a history of or concurrent neurological or psychiatric disease | RAVLT; BNT; TMT-B; CFT | HP<NC in the frequency of at least one e4 allele (8% vs. 26%) |

Table 1. Continued

| CITATION | TYPE OF OUTCOME | CHARACTERISTICS OF PARTICIPANTS (N) | DEFINITION OF THE HIGH-PERFORMING OLDER GROUP | OTHER INCLUSION CRITERIA | TESTS PERFORMED | MAIN OUTCOMES |
|----------------------------|-------------------|--|---|---|-------------------------------------|--|
| Gefen <i>et al.</i> (2014) | Cognitive profile | HP (18): mean age = 82.2 (2.4) <i>18-month follow-up</i> | Age ≥80 years Perform at or above average normative values for individuals in their 50s and 60s (RAVLT delayed-recall raw score ≥9) and within one standard deviation of the average for the non-memory measures | To have preserved activities of daily living and lacked clinical or structural evidence of a history of or concurrent neurological or psychiatric disease | RAVLT; BNT; TMT-B; CFT | HP did not show decline on memory, attention, language or executive function from baseline to 18 months. |
| Gefen <i>et al.</i> (2015) | Histology | HP (5): mean age = 88.6 (5.1), 5F, mean years of education = 17.2 (1.7) NC (5): mean age = 86.6 (8.6), 1M:4F, mean years of education = 13.8(2) | Age ≥80 years Perform at or above average normative values for individuals in their 50s and 60s (RAVLT delayed-recall raw score ≥9) and within one standard deviation of the average for the non-memory measures | To lack clinical evidence or history of neurologic or psychiatric disease | RAVLT; BNT; TMT-A; TMT-B; CFT; MMSE | Mean numerical estimates of Amyloid plaques and Neurofibrillary tangles density were lowest in HP. HP > YG=NC of Von Economo Neurons in anterior midcingulate cortex, in which neuron density was 3- to 5-fold higher in HP. |

Table 1. Continued

| CITATION | TYPE OF OUTCOME | CHARACTERISTICS OF PARTICIPANTS (N) | DEFINITION OF THE HIGH-PERFORMING OLDER GROUP | OTHER INCLUSION CRITERIA | TESTS PERFORMED | MAIN OUTCOMES |
|-----------------|-----------------------------|--|---|--|---|--|
| Mapstone (2016) | Plasma metabolites | HP (41): mean age = 83.2 (3.3), 20M:21F, mean years of education = 16.4 (2.6) NC (41): mean age = 83.2 (3.8), 20M:21F, mean years of education = 16.2 (2.4) | Age ≥ 70 years. Performed a composite memory Z-score > 1.35 SD. Other cognitive functions were required to be > -1.35 SD | To have good overall physical health, visual acuity and hearing sufficient for cognitive testing, proficiency in English language To lack major neurological or psychiatric illness, chronic abnormalities in blood count | RAVLT, FDS (of the WMS-III), TMT-A, TMT-B, BNT, CFT, HVOT | HP > NC in a 12-metabolites panel (Aspartate, Hydroxyhexa-decadienylcarnitine (C16:2-OH), 3-Hydroxy-pa-imitoleylcarnitine (C16:1-OH), Lyso PC a C28:1, Arginine, Valerylcarnitine (C5), Lyso PC a C17:0, Asparagine, Citrulline, Nitrotyrosine, PC aa C38:5, and Histamine). |
| Cook 2017 | Longitudinal Structural MRI | HP (24): mean age = 83.3 (3.5), 6M:18F, mean years of education = 15 (2.4) NC (12): mean age = 83.4 (3.8), 7M:5F, mean years of education = 15.6 (4.1) | Age ≥ 80 years Perform at or above average normative values for individuals in their 50s and 60s (RAVLT delayed-recall raw score ≥ 9) and within one standard deviation of the average for the non-memory measures | To have preserved activities of daily living and lacked clinical or structural evidence of a history of or concurrent neurological or psychiatric disease | RAVLT; BNT; TMT-B; CFT | HP < NC in annual percent change of whole-brain cortical volume loss (18 months apart). |

Table 1. Continued

| CITATION | TYPE OF OUTCOME | CHARACTERISTICS OF PARTICIPANTS (N) | DEFINITION OF THE HIGH-PERFORMING OLDER GROUP | OTHER INCLUSION CRITERIA | TESTS PERFORMED | MAIN OUTCOMES |
|-------------------------------|--|---|---|--|--|--|
| Dekhtyar <i>et al.</i> (2017) | Longitudinal Structural MRI Amyloid PET APOE pattern | HP (25): mean age = 77.5 (6.7), 9M:16F, mean years of education = 16 (6) NC (100): mean age = 78.89 (5.5), 47M:53F, mean years of education = 16 (5) | Age ≥ 75 years Memory Composite ≥ 0.5 SD. <i>Maintainers:</i> three-year follow-up with Memory Composite ≥ 0.5 SD | To have a normal score on the MMSE, Logical Memory II (of the WMS-R) and CDR. To have no history of alcoholism or drug abuse in the last two years, head trauma, or current serious medical or psychiatric illness | Memory composite: delayed scores of the MCT and FNAME. FAS, Letter-number of the WMS-III, DSB, Flanker, TMT-A, TMT-B minus A, Digit Symbol of the WAIS-R | HP > NC hippocampal volumes. HP = NC in level of amyloid burden. HP > NC in Composites of Executive functioning and Processing Speed <i>Maintainers:</i> HP = NC hippocampal volumes. HP < NC in level of amyloid burden HP < NC in the frequency of ε4 allele (16% vs. 30%) |

Table 1. Continued

| CITATION | TYPE OF OUTCOME | CHARACTERISTICS OF PARTICIPANTS (N) | DEFINITION OF THE HIGH-PERFORMING OLDER GROUP | OTHER INCLUSION CRITERIA | TESTS PERFORMED | MAIN OUTCOMES |
|------------------------------|-------------------------------|---|--|---|--|---|
| Janecek <i>et al.</i> (2017) | Acetylcholinesterase activity | HP (5): mean age = 90.2 (2.9), 5F NC (15): mean age = 83.3 (8), 9M:6F | Age ≥80 years. Perform at or above average normative values for individuals in their 50s and 60s (RAVLT delayed-recall raw score ≥ 9) and within one standard deviation of the average for the non-memory measures | To have no indication of ante mortem neurologic or psychiatric disorders | RAVLT; BNT; TMT-B; CFT. Careful chart review if neuro-psychological data not available | HP < NC staining intensity and density of acetylcholinesterase-positive cortical pyramidal neurons |
| Cook <i>et al.</i> (2017) | Psychological well-being | HP (31): median age = 83.4, 17M:23F NC (19): median age = 84.4, 7M:12F | Age ≥80 years. Perform at or above average normative values for individuals in their 50s and 60s (RAVLT delayed-recall raw score ≥ 9) and within one standard deviation of the average for the non-memory measures | To lack clinical evidence of significant neurologic or psychiatric illness. Maintain their cognitive status from enrollment to the time of questionnaires | RAVLT; BNT; TMT-B; CFT | HP > NC positive relations with others. HP = NC in other subscales of the well-being questionnaire (autonomy, environmental mastery, personal growth, purpose in life, self-acceptance) |

Note: HP – High-performing older adults, NC – Normal older controls, YG – Normal younger controls, MRI – Magnetic resonance imaging, RAVLT – Rey Auditory-Verbal Learning Test, BNT – Boston Naming Test, TMT – Trail making test, CFT – Category fluency test, MMSE – Mini-Mental State Examination, FDS – Forward Digit Span, WMS-III – Wechsler Memory Scale – 3rd edition, HVOT – Hooper Visual Organization test, PET – Positron Emission Tomography, MCT – Memory Capacity Test, FNAME – Face Name Associative Memory Exam, WMS-R – Wechsler Memory Scale – Revised; DSB – Digit Span Backwards, WAIS-R – Wechsler Adult Intelligence Scale – Revised, M:F – male:female

Table 2. Characteristics of included articles

| | HIGH-PERFORMING OLDER ADULTS | NORMAL OLDER CONTROLS |
|-------------------------------|---|-----------------------|
| Number of subjects (range) | 199 (5–41) | 548 (10–330) |
| Sex ratio | 47M:91F | 96M:105F |
| Mean age (years) | 82.5 | 73.9 |
| Minimum age (years) | 77.5 | 70 |
| Maximum age (years) | 90.2 | 83.7 |
| Mean education (years) | 13.6 | 16 |
| Outcome type (no. of studies) | 3 neuroimaging, 2 histology, 1 plasma metabolites, 2 neuropsychological profile, 1 ApoE | |
| Exclusion criteria | Samples including subjects with <70 years, lack of clear neuropsychological assessment, qualitative studies. | |
| Measure of cognitive profile | <p><i>Episodic memory:</i> Rey auditory-verbal learning test, delayed scores of the Memory Capacity Test and the Face Name Associative Memory Exam</p> <p><i>Other tests:</i> Logical Memory II, Backward and Forward Digit Span, Boston Naming Test, Trail Making Test (A, B, and A minus B), FAS, Category Fluency Test, Mini-Mental State Examination, Hooper Visual Organization Test, Digit Symbol Test, Flanker Test.</p> | |

ApoE – apolipoprotein E, M:F – male:female.

and the Face Name Associative Memory Exam. All included studies reported non-memory tests of the high-performing group similar to normal agers, usually fluency, naming, and attention skills. A longitudinal evaluation showed that most high-performing older adults exhibited no significant cognitive decline in memory and non-memory fields after 18 months of evaluation (Gefen *et al.*, 2014), but two individuals had lower memory scores at follow-up. Besides, this group showed higher level of positive social relationships when compared to age-matched controls, but both groups shared similar well-being score (Cook Maher *et al.*, 2017).

Neurobiological findings of high-performing older adults

Three studies evaluated high-performing older adults using neuroimaging techniques. Positron emission tomography (PET) was used in one paper, while magnetic resonance imaging was performed in all three studies; one had a cross-sectional design (Harrison *et al.*, 2012) and two used a longitudinal analysis with 18 month (Cook *et al.*, 2017) and three year follow-up (Dekhtyar *et al.*, 2017).

High-performing older adults showed global brain volume statistically indistinguishable from that of normal younger controls (average age = 57.9 years), and larger than that of normal older controls (average age = 83.1 years) (average whole-brain volume of High-performers vs. Older

controls = 288.05 vs. 244.13 mm³) (Harrison *et al.*, 2012). Moreover, the high-performing group showed increased thickness of left anterior cingulate (average thickness of High-performers vs. Older controls = 2.75 vs. 2.30 mm³), and increased hippocampal volumes in comparison to older controls (average volume of High-performers vs. Older Controls = 7,293 vs. 6,883 mm³) (Harrison *et al.*, 2012; Dekhtyar *et al.*, 2017). An 18-month follow-up showed an annual percent change of the whole-brain cortical volume loss significantly smaller in the SuperAgers group compared to normal older controls (annual percent change of High-performers vs. Older controls = 1.06% vs. 2.24%) (Cook *et al.*, 2017). A PET evaluation with PIB (*Pittsburgh Compound B*) was performed by Dekhtyar *et al.* (2017) and it revealed similar amyloid burden between the high-performing and normal older groups (median Distribution Volume Ratio or *DVR* of High-performers vs. Older controls = 1.16 vs. 1.11). In this same sample, all high-performing individuals whose scores did not decline within three years were classified as maintainers (16 of 25 individuals). This subgroup of maintainers showed lower amyloid burden at baseline compared to non-maintainers (Median *DVR* of maintainers vs. non-maintainers = 1.11 vs. 1.43), but both subgroups had similar hippocampal atrophy ($p = 0.850$) and amyloid accumulation ($p = 0.257$) rate over three years of follow-up assessment (Dekhtyar *et al.*, 2017).

Mapstone *et al.* (2017) analyzed the plasma metabolome of individuals with high memory capacity. The authors found a panel of 12 metabolites that could distinguish individuals with superior memory from controls, namely aspartate, hydroxyhexadecadienylcarnitine (C16:2-OH), 3-hydroxypalmitoleylcarnitine (C16:1-OH), lysophosphatidylcholine a C28:1, arginine, valerylcarnitine (C5), lysophosphatidylcholine a C17:0, asparagine, citrulline, nitrotyrosine, phosphatidylcholine aa C38:5, and histamine. Interestingly, an index developed with all 12 metabolites showed a significant relationship to a memory composite in the three studied groups. These metabolites also discriminated individuals with cognitive impairment from controls when their signs were reverted.

Two studies evaluated postmortem brain tissues of high-performing elderly individuals (Gefen *et al.*, 2015; Janeczek *et al.*, 2017). Gefen and colleagues reported the last cognitive evaluation of included individuals were within 24 months before death (range = 1–21 months). The authors showed that older adults with youthful memory scores had lower density of neurofibrillary tangles and amyloid plaques than controls in all cingulate areas, except the posterior midcingulate (Gefen *et al.*, 2015). Despite the lower density of pathological deposits, the high-performing group showed mixed Braak staging (from 0 to III). Besides, the anterior midcingulate had higher density of Von Economo neurons in the high-performing group compared to the other group. Total neuronal count and size were similar between the high-performing and control groups. Janeczek *et al.* (2017) evaluated five older adults with high memory performance for density and intensity of acetylcholinesterase (AChE) positivity in pyramidal neurons. They showed significantly lower density of AChE-positive neurons compared to older and younger controls in four described areas, namely the supplementary motor cortex, middle frontal gyrus, middle temporal gyrus, and inferior parietal lobe. The anterior cingulate cortex did not show statistical significance, despite the tendency of decreased density of AChE-positive neurons in the SuperAgers group. The high-performing group also showed decreased intensity in the middle frontal gyrus and middle temporal gyrus in comparison to older controls.

Genotyping for ApoE was described in three studies (Rogalski *et al.*, 2013; Dekhtyar *et al.*, 2017; Mapstone *et al.*, 2017). Rogalski *et al.* found that the high-performing older group had lower frequency of at least one e4 allele than that seen in normal controls (8% vs. 26%), while the other two studies found no statistically significant differences (16% vs. 30% and 12% vs. 9%).

Discussion

To our knowledge, this is the first review evaluating literature findings of high-performing older adults. Here, we described structural and molecular brain characteristics of individuals at 70 years of age or older with high memory performance compared to age-matched peers. While several studies have focused on successful aging, this review retrieved only studies regarding older adults with superior cognitive performance compared to their cognitively average peers. To select this specific sample, we included all studies that analyzed individuals with memory score of at least one standard deviation above average.

An operationalized definition of high-performing older adults is vital for the generalization of results, including age, cognitive measures, and study design. The age restriction for this review was based on previous studies that related an average onset of age-related memory decline at approximately 60–65 years of age (Rönnlund *et al.*, 2005; Schaie, 2005; Nyberg *et al.*, 2012). We considered 70 years of age an adequate, but not perfect cut-off. A lower limit of age would introduce a bias, while a higher limit would be too restrictive, as aging is a major risk factor for memory decline. Interestingly, episodic memory was measured in all included papers most of them (8/9 studies) used the Rey Auditory-Verbal Learning Test, though episodic memory evaluation was not an inclusion criterion. Typically, episodic memory shows a progressive decrease during the lifespan and it appears particularly vulnerable to aging (Hedden and Gabrieli, 2004; Harada *et al.*, 2013). Episodic memory evaluation at a single point is not a guarantee of cognitive maintenance, as in some high-performers may decline over time (Gefen *et al.*, 2014; Dekhtyar *et al.*, 2017). Non-memory measures were within the age-appropriate average in all included studies. Most studies compared the high-performing group to normal agers, except Harrison that also compared them to a middle-aged group (Harrison *et al.*, 2012). As mentioned by Nyberg *et al.* (2012), older adults with high performance may exhibit a more youthful brain phenotype. Thus, cognitive preservation is better evaluated with longitudinal studies. Moreover, a younger control group may provide important information on brain maintenance, possibly revealing subsequent mechanisms that may replicate memory preservation during senescence.

Despite the small number of studies on older adults with high cognitive performance, this group showed unique structural and molecular features when compared to normal agers. Structural findings of included studies suggest that

excellent memory ability is associated with global preservation of the cortex and decreased age-related atrophy, but it is not related to neuronal size or total count when compared to normal older controls (Harrison *et al.*, 2012; Gefen *et al.*, 2015; Cook *et al.*, 2017; Dekhtyar *et al.*, 2017). These alterations are in accordance with the brain maintenance view, but not with the brain reserve conception. Despite the hippocampal volumes were larger in high performers compared with normal performers, the hippocampal volumes and atrophy rates were similar in three years of follow-up between maintainers and non-maintainers. This finding suggests that the hippocampus is associated with the memory performance, but not with memory maintenance. At a molecular level, high-performing older adults showed lower levels of AD pathology when compared with older adults that showed a decrease in cognitive ability. Despite amyloid accumulation being similar between high-performing older adults and normal controls after three years, those that maintained an exceptional memory ability exhibited lower amyloid deposition at baseline. Neurofibrillary tangles and amyloid plaques were less present in histologic analysis of this group, especially in the anterior cingulate cortex. Moreover, high-performing older adults presented decreased acetylcholinesterase activity in a few brain regions, in contrast to the increase of this enzyme typically seen in age-related cognitive decline (Ashare *et al.*, 2012). Also, plasma metabolites successfully distinguished the high-performing older group from normal agers, indicating peripheral alterations associated with cognitive preservation. Among all metabolites significantly increased in this group, a few were associated with neuroplasticity and cognitive reserve, such as aspartate and NO (Schuman and Madison, 1991; Shimizu *et al.*, 2000; Nikonenko *et al.*, 2013). Consistent with the definition of brain maintenance, these findings suggest that lesser density of age-related lesions is related to better cognition in later life (de Frias *et al.*, 2007).

As proposed by Nyberg *et al.* (2012), structural and molecular preservation may mechanistically impact cognitive functioning. Combined, the findings of included studies on high-performing older adults may provide evidence toward a better understanding of cognitive aging. The maintenance of brain structures shown here may rely upon the marked similarity between brain structures of exceptional agers and younger adults, which are significantly thicker than those of typical older adults (Salthouse, 2009). The persistence of high performance in older adults may result from mitigating neurobiological errors by mechanisms yet to be identified, probably

associated with neuroplasticity (Heuninckx *et al.*, 2008; Barulli and Stern, 2013). The avoidance of amyloid pathology, as showed by the subgroup of maintainers (Dekhtyar *et al.*, 2017), may lead to decreased neurodegeneration and consequently higher cognitive functioning. It is putative that both the reserve and maintenance theories converge as complementary concepts (Barulli and Stern, 2013; Habeck *et al.*, 2016). As the adult lifespan is marked by greater cognitive enrichment, the cognitive reserve of high-performing older adults could protect against impairment by reducing age-related pathology to the established networks in older life (Sumowski *et al.*, 2010). However, both reserve concepts do not cover the preservation of cognitive abilities during the aging process (Habeck *et al.*, 2016). However, the current body of literature is insufficient to offer a solid conclusion, as few studies have adequately addressed this group.

Additionally, tau pathology is strongly associated with memory impairment (Riley *et al.*, 2002; Braak *et al.*, 2006). As a single study was inconclusive on tau pathology in autopsies of high-performing older adults, future studies should target tau imaging in this group. Several studies using fMRI have indicated that individuals with age-related cognitive decline rely on compensatory brain activity to preserve function-specific memory networks (Cabeza, 2002; Davis *et al.*, 2008; Park and Reuter-Lorenz, 2009; O'Brien *et al.*, 2010; Eyler *et al.*, 2011). Functional connectivity of high-performing older adults remains unclear, but its elucidation is essential in order to determine the optimal functioning of established neural networks. Both techniques hold great promise in solving the aging brain puzzle.

The risk of biases must be discussed. Despite our efforts, some important papers may have been omitted due to a lack of consensus on the definition of successful aging (Depp *et al.*, 2010; Depp *et al.*, 2011). Further, some studies were not controlled for basic variables, such as sex, especially those including histologic analyses. The total number of studies and the heterogeneity of their results may hinder the generalization of our findings. We performed a comprehensive search with almost no factor of limitation to minimize this bias, but seven of nine included studies were from the same group. Meta-analysis was not possible due to the restricted number of papers on this subject and their heterogeneity of existing papers. Cross-sectional studies are influenced by cohort effects, which can overestimate the study's findings. An estimated prevalence of high-performers is limited in this work because of the design of included studies. Finally, our conclusions may be affected by the small number of studies and its limitations.

In sum, this review draws attention to the study of high-performing, rather than simply healthy, older adults. Despite the insufficient number of studies to draw a consistent conclusion, the compliance of findings in this work corroborates the concept of brain maintenance. High-performing older adults exhibited particular structural and molecular characteristics, such as a preserved cortical volume and decreased AD pathology in the brain. As only few studies provided clear, objective definition criteria for high-performing older adults, further longitudinal investigations with younger controls are necessary to reach concrete conclusions.

Conflict of interest

The authors declare that they have no conflict of interest.

Author contributions

JCC coordinated, designed, and revised this study. WVB designed, analyzed, and contributed in the writing of the manuscript, and the screening of the studies. LP and LBF contributed in the methodology and the screening of the studies. GR contributed with methodological aspects. MWP and LPS contributed to the writing of the manuscript and the review of this study.

Acknowledgments

None.

References

- Ashare, R. L., Ray, R., Lerman, C. and Strasser, A. A. (2012). Cognitive effects of the acetylcholinesterase inhibitor, donepezil, in healthy, non-treatment seeking smokers: a pilot feasibility study. *Drug and Alcohol Dependence*, 126, 263–267. doi: 10.1016/j.drugalcdep.2012.04.019.
- Barulli, D. and Stern, Y. (2013). Efficiency, capacity, compensation, maintenance, plasticity: emerging concepts in cognitive reserve. *Trends in Cognitive Sciences*, 17, 502–509. doi: 10.1016/j.tics.2013.08.012.
- Braak, H., Alafuzoff, I., Arzberger, T., Kretschmar, H. and Del Tredici, K. (2006). Staging of Alzheimer disease-associated neurofibrillary pathology using paraffin sections and immunocytochemistry. *Acta Neuropathologica*, 112, 389–404. doi: 10.1007/s00401-006-0127-z.
- Cabeza, R. (2002). Hemispheric asymmetry reduction in older adults: the HAROLD model. *Psychology and Aging*, 17, 85–100. doi: 10.1037//0882-7974.17.1.85.
- Cook Maher, A. *et al.* (2017). Psychological well-being in elderly adults with extraordinary episodic memory. *Plos One*, 12, e0186413. doi: 10.1371/journal.pone.0186413.
- Cook, A. H. *et al.* (2017). Rates of cortical atrophy in adults 80 years and older with superior vs average episodic memory. *JAMA*, 317, 1373. doi: 10.1001/jama.2017.0627.
- Cummings, J. L., Morstorf, T. and Zhong, K. (2014). Alzheimer's disease drug-development pipeline: few candidates, frequent failures. *Alzheimer's Research & Therapy*, 6, 37. doi: 10.1186/alzrt269.
- Davis, S. W., Dennis, N. A., Daselaar, S. M., Fleck, M. S. and Cabeza, R. (2008). Que PASA? The posterior-anterior shift in aging. *Cerebral Cortex*, 18, 1201–1209. doi: 10.1093/cercor/bhm155.
- de Frias, C. M., Lövdén, M., Lindenberger, U. and Nilsson, L.-G. (2007). Revisiting the dedifferentiation hypothesis with longitudinal multi-cohort data. *Intelligence*, 35, 381–392. doi: 10.1016/j.intell.2006.07.011.
- Dekhtyar, M. *et al.* (2017). Neuroimaging markers associated with maintenance of optimal memory performance in late-life. *Neuropsychologia*, 100, 164–170. doi: 10.1016/j.neuropsychologia.2017.04.037.
- Depp, C. A., Harmell, A. and Vahia, I. V. (2011). Successful cognitive aging. *Current Topics in Behavioral Neurosciences*, 10, 35–50. doi: 10.1007/7854_2011_158.
- Depp, C., Vahia, I. and Jeste, D. (2010). Successful aging: focus on cognitive and emotional health. *Annual Review of Clinical Psychology*, 6, 527–550.
- Eyler, L. T., Sherzai, A., Kaup, A. R. and Jeste, D. V. (2011). A review of functional brain imaging correlates of successful cognitive aging. *Biological Psychiatry*, 70, 115–122. doi: 10.1016/j.biopsych.2010.12.032.
- Gefen, T. *et al.* (2014). Longitudinal neuropsychological performance of cognitive SuperAgers. *Journal of the American Geriatrics Society*, 62, 1598–600. doi: 10.1111/jgs.12967.
- Gefen, T. *et al.* (2015). Morphometric and histologic substrates of cingulate integrity in elders with exceptional memory capacity. *Journal of Neuroscience*, 35, 1781–1791. doi: 10.1523/JNEUROSCI.2998-14.2015.
- Habeck, C., Razlighi, Q., Gazes, Y., Barulli, D., Steffener, J. and Stern, Y. (2016). Cognitive reserve and brain maintenance: orthogonal concepts in theory and practice. *Cerebral Cortex*, 27, 3962–3969. doi: 10.1093/cercor/bhw208.
- Harada, C. N., Natelson Love, M. C. and Triebel, K. L. (2013). Normal cognitive aging. *Clinics in Geriatric Medicine*, 29, 737–752. doi: 10.1016/j.cger.2013.07.002.
- Harrison, T. M., Weintraub, S., Mesulam, M.-M. M.-M. and Rogalski, E. (2012). Superior memory and higher cortical volumes in unusually successful cognitive aging. *Journal of the International Neuropsychological Society*, 18, 1081–1085. doi: 10.1017/S1355617712000847.
- Hedden, T. and Gabrieli, J. D. E. (2004). Insights into the ageing mind: a view from cognitive neuroscience. *Nature Reviews Neuroscience*, 5, 87–96. doi: 10.1038/nrn1323.
- Heuninckx, S., Wenderoth, N. and Swinnen, S. P. (2008). Systems neuroplasticity in the aging brain: recruiting additional neural resources for successful motor performance in elderly persons. *Journal of Neuroscience*, 28, 91–99. doi: 10.1523/JNEUROSCI.3300-07.2008.

- Janeczek, M. et al.** (2017). Variations in acetylcholinesterase activity within human cortical pyramidal neurons across age and cognitive trajectories. *Cerebral Cortex*, 1–9. doi: 10.1093/cercor/bhx047.
- Mapstone, M. et al.** (2017). What success can teach us about failure: the plasma metabolome of older adults with superior memory and lessons for Alzheimer's disease. *Neurobiology of Aging*, 51, 148–155. doi: 10.1016/j.neurobiolaging.2016.11.007.
- Moher, D., Liberati, A., Tetzlaff, J. and Altman, D. G.** (2009). Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *BMJ*, 339, b2535–b2535. doi: 10.1136/bmj.b2535.
- Nikonenko, I., Nikonenko, A., Mendez, P., Michurina, T. V., Enikolopov, G. and Muller, D.** (2013). Nitric oxide mediates local activity-dependent excitatory synapse development. *Proceedings of the National Academy of Sciences of the United States of America*, 110, E4142–E4151. doi: 10.1073/pnas.1311927110.
- Nyberg, L., Lövdén, M., Riklund, K., Lindenberger, U. and Bäckman, L.** (2012). Memory aging and brain maintenance. *Trends in Cognitive Sciences*, 16, 292–305. doi: 10.1016/j.tics.2012.04.005.
- O'Brien, J. L. et al.** (2010). Longitudinal fMRI in elderly reveals loss of hippocampal activation with clinical decline. *Neurology*, 74, 1969–1976. doi: 10.1212/WNL.0b013e3181e3966e.
- Park, D. C. and Reuter-Lorenz, P.** (2009). The adaptive brain: aging and neurocognitive scaffolding. *Annual Review of Psychology*, 60, 173–196. doi: 10.1146/annurev.psych.59.103006.093656.
- Prince, M., Wimo, A., Guerchet, M., Gemma-Claire, A., Wu, Y.-T. and Prina, M.** (2015). *World Alzheimer Report 2015: The Global Impact of Dementia – An Analysis of Prevalence, Incidence, Cost and Trends*. London: Alzheimer's Disease International, 84. doi: 10.1111/j.0963-7214.2004.00293.x.
- Riley, K. P., Snowdon, D. A. and Markesbery, W. R.** (2002). Alzheimer's neurofibrillary pathology and the spectrum of cognitive function: findings from the nun study. *Annals of Neurology*, 51, 567–577. doi: 10.1002/ana.10161.
- Rogalski, E. J. et al.** (2013). Youthful memory capacity in old brains: anatomic and genetic clues from the northwestern superaging project. *Journal of Cognitive Neuroscience*, 25, 29–36. doi: 10.1162/jocn_a_00300.
- Rönnlund, M., Nyberg, L., Bäckman, L. and Nilsson, L.-G.** (2005). Stability, growth, and decline in adult life span development of declarative memory: cross-sectional and longitudinal data from a population-based study. *Psychology and Aging*, 20, 3–18. doi: 10.1037/0882-7974.20.1.3.
- Salthouse, T. A.** (2009). When does age-related cognitive decline begin? *Neurobiology of Aging*, 30, 507–514. doi: 10.1016/j.neurobiolaging.2008.09.023.
- Schaie, K. W.** (2005). *Developmental Influences on Adult Intelligence: The Seattle Longitudinal Study*. New York: Oxford University Press. doi: 10.1093/acprof:oso/9780195156737.001.0001.
- Schuman, E. M. and Madison, D. V.** (1991). A requirement for the intercellular messenger nitric oxide in long-term potentiation. *Science*, 254, 1503–1506. Available at: <http://www.ncbi.nlm.nih.gov/pubmed/1720572>.
- Shimizu, E., Tang, Y. P., Rampon, C. and Tsien, J. Z.** (2000). NMDA receptor-dependent synaptic reinforcement as a crucial process for memory consolidation. *Science*, 290, 1170–1174. Available at: <http://www.ncbi.nlm.nih.gov/pubmed/11073458>.
- Stern, Y.** (2009). Cognitive reserve☆. *Neuropsychologia*, 47, 2015–2028. doi: 10.1016/j.neuropsychologia.2009.03.004.
- Sumowski, J. F., Wylie, G. R., DeLuca, J. and Chiaravalloti, N.** (2010). Intellectual enrichment is linked to cerebral efficiency in multiple sclerosis: functional magnetic resonance imaging evidence for cognitive reserve. *Brain*, 133, 362–374. doi: 10.1093/brain/awp307.
- Sun, F. W., Stepanovic, M. R., Andreano, J., Barrett, L. F., Touroutoglou, A. and Dickerson, B. C.** (2016). Youthful brains in older adults: preserved neuroanatomy in the default mode and salience networks contributes to youthful memory in superaging. *The Journal of Neuroscience: The Official Journal of the Society for Neuroscience*, 36, 9659–9668. doi: 10.1523/JNEUROSCI.1492-16.2016.

Save the Date

IPA/SEPG Joint
International Congress



31 AUGUST - 3 SEPTEMBER 2019



Santiago de Compostela, Spain



International Psychogeriatric Association
Better Mental Health for Older People

SEPG

Sociedad Española
de Psicogeriatría

www.ipa-online.org/2019congress

Cardiovascular health and cognitive function among Mexican older adults: cross-sectional results from the WHO Study on Global Ageing and Adult Health

Jaime Perales,¹ Ladson Hinton,² Jeffrey Burns¹ and Eric D. Vidoni¹

¹Department of Neurology, University of Kansas Alzheimer's Disease Center, Fairway, Kansas, USA

²Department of Psychiatry and Behavioral Sciences, University of California, Davis, California, USA

ABSTRACT

Objectives: To assess the association between cardiovascular health and cognitive function among Mexican older adults.

Design: Nationally representative cross-sectional survey.

Setting: Households in Mexico.

Participants: Individuals aged 50 years and older ($n = 1,492$) from the Mexico-SAGE project Wave 1.

Measurements: A continuous and a categorical index of cardiovascular health was calculated based on exercise, smoking, body mass index, and blood pressure ranging from 0 to 4. Cognitive function was obtained by averaging the standardized scores (z scores) of five psychometric tests. Associations were conducted using linear regression.

Results: The continuous index of cardiovascular health was not associated with cognitive function. Using the categorical index, participants with the best levels of cardiovascular (score of 4) health performed better on global cognitive function than groups with lower cardiovascular health (scores of 0, 0.41 SD ; 1, 0.39 SD ; and 2, 0.56 SD). The association was moderated by age, reaching significance only among those 50–64 years old.

Conclusions: If longitudinal research confirms these findings, results would suggest that dementia-related policies in Mexico need to focus on achieving optimal levels of cardiovascular health, especially in midlife.

Key words: cognitive activity, dementia, aging

Introduction

Dementia and cognitive decline are among the greatest public health challenges Mexico will face in the coming years. Mexico's population has quickly aged in the last decades. Mexicans 60 years and older will represent one in every five people by 2030 (Instituto Nacional de Salud Pública [INSP], 2014). Meanwhile, the average life expectancy, currently 77, continues to increase to a projected 80 by 2050 (Instituto Nacional de Salud Pública [INSP], 2014; World Health Organization, 2015). This population shift has led to one of the highest rates of dementia in the Americas, currently 30.4 per 1,000 person-years among Mexicans 65

years and older (Prince *et al.*, 2013). A better understanding of brain health is needed to reduce the health and financial impact of dementia on individuals, families, and societies (World Health Organization, 2012).

Studies such as the Mexican Health and Aging study show that individual cardiovascular risk factors are associated with cognitive decline and dementia (Mejía-Arango *et al.*, 2007; Mejía-Arango and Gutierrez, 2011; Silvia and Clemente, 2011). Cardiovascular risk indices have been associated with cognition among Mexican Americans in the Sacramento Area Latino Study on Aging study and other populations (Yaffe *et al.*, 2007; Unverzagt *et al.*, 2011; Al Hazzouri *et al.*, 2013). Health indices of factors that coexist and have a common pathway are promising as they are more comprehensive than single factors (Pearson *et al.*, 2013). However, prevention strategies need to also focus on optimal levels of modifiable health to increase population impact rather than merely on poor levels (Lloyd-Jones *et al.*, 2010).

Correspondence should be addressed to: Eric D. Vidoni, Department of Neurology, University of Kansas Alzheimer's Disease Center, MS6002, Fairway, Kansas 66205, USA. Phone: +1-913-588-5312. Email: evidoni@kumc.edu. Received 6 Jan 2018; revision requested 28 Jan 2018; revised version received 8 Feb 2018; accepted 13 Feb 2018. First published online 18 April 2018.

The American Heart Association (AHA) defined a cardiovascular health (CVH) index to track health status in relation to their 2020 strategic goal (Lloyd-Jones *et al.*, 2010). This concept includes a set of modifiable cardiovascular risk factors and health behaviors graded either poor, intermediate, or ideal. The AHA's CVH index has been shown to be associated with lower incidence of stroke, cardiovascular disease, and related mortality (Dong *et al.*, 2012; Kulshreshtha *et al.*, 2013). A few studies in the USA have found associations between the AHA's CVH index and cognitive outcomes among people of different ethnic backgrounds (Reis *et al.*, 2013; Crichton *et al.*, 2014; Thacker *et al.*, 2014; Gardener *et al.*, 2016; González *et al.*, 2016). For example, one cross-sectional study found positive associations between CVH and scores of cognitive status, verbal learning, phonemic word fluency, and processing speed among USA middle-aged Latinos (González *et al.*, 2016). The Northern Manhattan Study found that in a sample with 65% of mostly Caribbean Latinos, the higher number of ideal CVH metrics was associated with less decline in different cognitive domains (Gardener *et al.*, 2016).

Determining the association between CVH and cognitive function in Mexico is important to promote dementia and cognitive decline prevention and healthy brain aging in this country. Mexico has not just one of the highest rates of dementia in the Americas but also the highest prevalence of diabetes (Shaw *et al.*, 2010), obesity (World Obesity Federation, 2017), and hypertension (Ordúñez *et al.*, 2001). Therefore, the aim of this study is to explore the association between an index of ideal levels of modifiable CVH factors and cognitive function among Mexican older adults using nationally representative cross-sectional data. We hypothesize that better levels of CVH will be associated with higher cognitive function. Previous research shows that some CVH components including body mass index (BMI) are associated with dementia in midlife but not later in life (Fitzpatrick *et al.*, 2009). This study also aimed to explore whether the association CVH and cognitive function is moderated by age. This analysis builds on prior research in Mexico by using an index of ideal levels of modifiable CVH factors as opposed to individual risk factors or indexes that include non-modifiable factors.

Methods

The current study used cross-sectional data from Wave 1 of the World Health Organization Study on Global Ageing and Adult Health (SAGE)

undertaken in Mexico. This is an observational, longitudinal study representative of the general non-institutionalized adult population (18 years or older).

Sample and procedure

The methodology for SAGE has been published elsewhere (Kowal *et al.*, 2012). In brief, the survey was conducted between 2009 and 2010. A stratified, multi-stage cluster sampling design was used to obtain a nationally representative sample. A probability proportion to size design was used to select clusters. Within each cluster, an enumeration of existing households was done to obtain an accurate measurement of size. As the focus of SAGE was on older adults, individuals aged ≥ 50 years were oversampled. Interviews were conducted face-to-face at respondent's homes using Paper and Pencil Assisted Interview (PAPI). All the interviewers participated in a training course for the administration of the survey protocol. Quality control procedures were implemented during fieldwork (Üstun *et al.*, 2005). Those participants who were not able to respond to the survey due to cognitive problems were administered a proxy interview. The individual response rate was 51%. Sampling weights were generated to account for the sampling design. Post-stratification corrections were made to the weights to adjust for the population distribution obtained from the national census and for non-response.

Ethical approvals followed the ethical guidelines of the 1975 Declaration of Helsinki and were obtained from the Institutional Review Board of the National Institute of Public Health, Cuernavaca, Mexico. Study procedures for the secondary analysis were supervised by the University of Kansas Medical Center's Institutional Review Board as not involving human subjects research. Written informed consent from each participant was obtained by the National Institute of Public Health, Cuernavaca, Mexico. The SAGE dataset is publicly available at the WHO website (<http://apps.who.int/healthinfo/systems/surveydata/index.php/catalog/sage>).

Measures

Cognitive function

Cognitive function was assessed using performance Spanish language measures of immediate and delayed verbal recall, verbal fluency, and forward and backward digit span. Verbal memory was tested using the Consortium to Establish a Registry for Alzheimer's disease verbal recall test, consisting of

three repetitions of a ten-word list for immediate recall with scores ranging from 0 to 30 words recalled correctly, and then assessing the recall of these words after a 10-minute delay with scores ranging from 0 to 10 words recalled correctly (Morris *et al.*, 1988). The verbal fluency test consisted of naming as many animals as possible in 1 minute with scores being the sum of different words correctly named (Morris *et al.*, 1988). Digit span was used to assess memory capacity and executive function, using both forward and backward digit recall tests (Wechsler *et al.*, 1997). The forward digit span version contained nine and the backward version eight series. Each series had two trials. The score in each digit span task was the series number in the longest series repeated without error in the first or second trial. An overall cognitive function score was calculated by averaging the standardized scores of each test. All cognitive tests have been validated in the Mexican population (Ostrosky-Solis *et al.*, 1999; Wechsler, 2001; Tulsy and Zhu, 2003; Escobedo and Hollingworth, 2009; Sosa *et al.*, 2009). The score in each digit span task was the series number in the longest series repeated without error in the first or second trial. An overall cognitive function score was calculated by averaging the standardized scores of each test.

Cardiovascular health index

We obtained the CVH indices following four of the seven criteria from the AHA definition (Lloyd-Jones *et al.*, 2010). Full data on the remaining three criteria (fasting cholesterol, glucose, and diet) was not collected in the survey, and therefore was not included in the composite measures. All participants were asked whether they had ever used tobacco. Participants who had used tobacco were asked whether they currently used it daily, non-daily, or not at all. Those who reported former tobacco use were asked how old they were when they stopped using tobacco. Participants who had never smoked or quit more than 12 months before the survey were considered to have ideal smoking status in that criterion. Those who were current smokers or quit within the last 12 months were considered to have poor/intermediate smoking status. The Global Physical Activity Questionnaire (GPAQ) was used to measure the intensity, duration, and frequency of physical activity in three domains: occupational; transport-related; and discretionary or leisure time (Bull *et al.*, 2009). The Spanish GPAQ has been validated among an almost fully Mexico-born sample in California (Hoos *et al.*, 2012). The total time spent in physical activity during a typical week, including the number of days and intensity, were used to generate categories

of ideal physical activity levels (≥ 150 minutes per week of moderate intensity, ≥ 75 minutes per week of vigorous intensity, or a combination of both) and poor/intermediate levels (0–149 minutes per week of moderate intensity, 0–74 minutes per week of vigorous intensity or a combination of both). Weight and height were measured to calculate BMI, calculated as weight/height² (kg/m²). Participants with a BMI < 25 kg/m² were considered to have ideal BMI levels and those with higher BMI were considered to have poor/immediate levels. Blood pressure was measured three times on the right arm of the seated respondent using a wrist blood pressure monitor. Out of three measurements, an average of the first two measurements was used as the blood pressure value in this analysis. Participants with systolic values < 120 and diastolic values < 80 mmHg were considered to have ideal blood pressure levels. Those with either systolic or diastolic blood pressure levels than ideal were considered to have poor/immediate levels. The four criteria for ideal CVH are in line with the Mexican national guidelines for tobacco use, physical activity, BMI, and blood pressure (Secretaría de Salud, 1999, 2009; Bonvecchio *et al.*, 2015). Participants obtained a score of 1 if they met the ideal criterion for each component (smoking, physical activity, BMI, and blood pressure) and a 0 otherwise, with total score ranging from 0 to 4 points (Table 1). We used two definitions of CVH, first as a continuous variable and second as a categorical variable with five groups (0, 1, 2, 3, and 4). Secondary analyses examined the moderating effect of age (50–64 years and 65+ years) in the association between CVH and cognitive function and the association of cognition with each of the CVH components individually.

Covariates

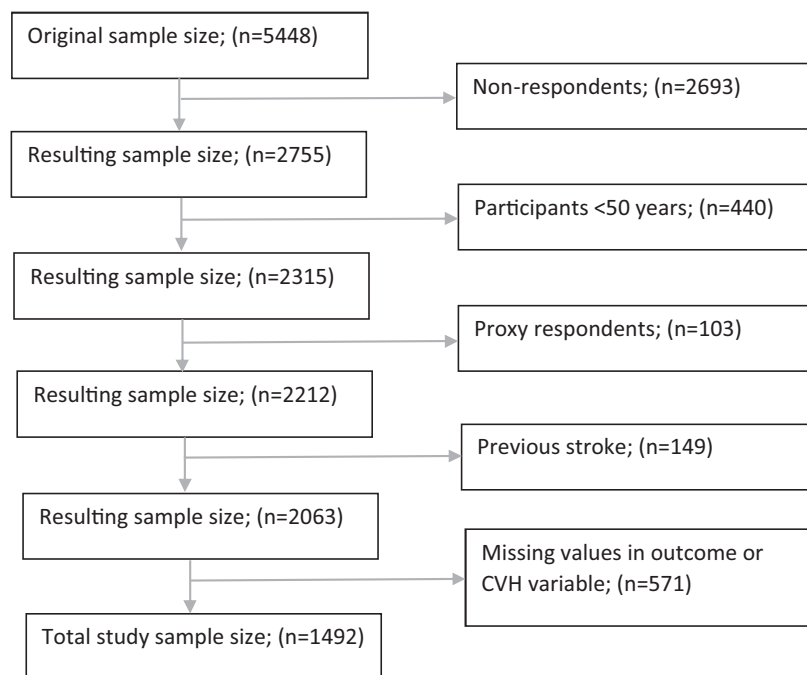
Socio-demographic information included age (continuous), gender, years of education (continuous), wealth (continuous), and urbanicity (rural/urban). For wealth, a multi-step process was used where asset ownership was first converted to an asset ladder, and then the Bayesian post-estimation method was used to generate raw continuous income estimates. Urbanicity was defined consistently with the way the areas were legally proclaimed to be, including towns, cities and metropolitan areas (urban), commercial farms, small settlements, rural villages, and areas further away from towns and cities (rural).

Statistical methods

The analysis was restricted to participants aged ≥ 50 years. Proxy respondent data was not included

Table 1. Criteria for ideal or poor/intermediate levels of cardiovascular health of individual factors according to the index

| CVH INDEX | POOR/INTERMEDIATE | IDEAL |
|-------------------|---|---|
| | (0 points) | (1 point) |
| Smoking status | Current smoker or quit ≤ 12 months ago | Never smoker or quit > 12 months ago |
| Physical activity | 0–149 minute/week moderate intensity or 0–74 minute/week vigorous intensity | ≥ 150 minute/week moderate intensity or ≥ 75 minute/week vigorous intensity |
| Body mass index | ≥ 25 kg/m ² | < 25 kg/m ² |
| Blood pressure | SBP ≥ 120 or DPB ≥ 80 mmHg | SBP < 120 and DPB < 80 mmHg |

**Figure 1.** (Colour online) Sample flowchart and reasons for exclusion.

as they did not provide information on key variables. Participants with stroke assessed through either an algorithm or self-report of diagnosis were excluded from the sample as the association of CVH and stroke is well established (Dong *et al.*, 2012; Kulshreshtha *et al.*, 2013). Descriptive analyses included weighted percentages, unweighted frequencies, means, and standard errors. Models controlled for age, gender, education, wealth, and urbanicity. Interaction analysis was conducted to assess the moderating effect of age. Cross-sectional analyses were conducted using linear regression. The level for statistical significance for all analyses was set at $p < 0.05$. Complete case analysis was conducted. Figure 1 shows a flowchart with information on how the final sample was reached. Participants deleted based on missing values in CVH or cognitive function ($n = 571$) did not differ statistically from those not missing in age, gender,

education, wealth, and urbanicity. All analyses were performed using SPSS version 22.0 using complex samples analysis (IBM Corp. Released, 2013).

Results

Among the 1,492 participants included in this analysis, the mean age was 61.6 years, ranging from 50 to 93 years and 53.6% were women. The average years of education was 5.1 and 21.6% lived in a rural setting (see Table 2). Socio-demographic factors were not associated with CVH. CVH was worst (score 0) for 8.4%, and best (score 4) for 2.2%.

Table 3 shows the bivariate and multivariate linear regressions of global cognitive function with the continuous and categorical CVH indices and

Table 2. Descriptive statistics of SAGE participants aged 50 years and over by cardiovascular health (CVH)

| | TOTAL SAMPLE; N = 1,492 | CVH 0; N = 105 (8.4%) | CVH 1; N = 607 (36.0%) | CVH 2; N = 576 (40.2%) | CVH 3; N = 178 (13.2%) | CVH 4; N = 26 (2.2%) | P VALUE |
|--------------------------|----------------------------|-----------------------------|------------------------------|------------------------------|------------------------------|----------------------------|---------|
| Age, mean (SE) | 61.6 (1.0) | 61.7 (1.2) | 61.5 (1.4) | 61.9 (1.3) | 61.8 (3.0) | 57.0 (2.7) | 0.85 |
| Age, 50–64; n (%) | 669 (69.1) | 41 (70.9) | 273 (70.3) | 260 (65.5) | 81 (73.7) | 14 (82.4) | 0.89 |
| Gender, women; n (%) | 936 (53.6) | 40 (42.3) | 396 (44.8) | 372 (58.5) | 115 (75.0) | 13 (25.3) | 0.06 |
| Education, mean (SE) | 5.9 (0.5) | 5.5 (0.7) | 5.7 (0.6) | 4.9 (1.3) | 3.8 (1.7) | 4.3 (1.3) | 0.86 |
| Wealth, mean (SE) | 0.1 (0.0) | 0.1 (0.1) | 0.1 (0.1) | 0.1 (0.1) | 0.1 (0.1) | 0.1 (0.1) | 0.79 |
| Urbanicity, rural; n (%) | 403 (21.6) | 18 (6.3) | 153 (28.4) | 166 (18.8) | 58 (14.2) | 8 (64.3) | 0.80 |

Table 3. Relationship of continuous and categorical indices of CVH and covariates with global cognitive function in the total sample and by age group

| | TOTAL SAMPLE | | AGES 50–64 | AGES 65+ |
|-------------------------------|--|---|--|--|
| | BIVARIATE LINEAR REGRESSION ^a | MULTIVARIATE LINEAR REGRESSION WITH CONTINUOUS CVH ^b | MULTIVARIATE LINEAR REGRESSION WITH CATEGORICAL CVH ^b | MULTIVARIATE LINEAR REGRESSION WITH CATEGORICAL CVH ^c |
| CVH (continuous) | 0.00 (−0.11;0.11) | 0.02 (−0.56;0.10) | – | – |
| CVH 4 | | | | |
| CVH 3 | −0.28 (−0.73;0.18) | – | −0.29 (−0.64;0.06) | −0.42 (−0.83;−0.01) |
| CVH 2 | −0.47 (−0.83;−0.10) | – | −0.56 (−0.88;−0.25) | −0.68 (−1.04;−0.32) |
| CVH 1 | −0.31 (−0.69;0.07) | – | −0.39 (−0.73;−0.05) | −0.49 (−0.85;−0.12) |
| CVH 0 | −0.31 (−0.69;0.07) | – | −0.41 (−0.77;−0.05) | −0.56 (−0.97;−0.14) |
| Gender; women (men REF) | −0.11 (−0.34;0.12) | −0.04 (−0.23;0.15) | −0.02 (−0.18;0.13) | 0.01 (−0.18;0.20) |
| Age (per unit) | −0.03 (−0.03;−0.02) | −0.02 (−0.27;−0.01) | −0.02 (−0.03;−0.01) | – |
| Education (per unit) | 0.05 (0.04;0.07) | 0.03 (0.01;0.05) | 0.03 (0.02;0.04) | 0.03 (0.01;0.05) |
| Wealth (per unit) | 0.74 (0.61;0.86) | 0.40 (0.19;0.62) | 0.39 (0.21;0.57) | 0.38 (0.17;0.58) |
| Urbanicity; urban (rural REF) | 0.36 (0.20;0.53) | 0.14 (−0.04;0.33) | 0.18 (0.00;0.35) | 0.25 (0.03;0.48) |

^aBivariate associations between each of the variables in the column and global cognitive function independently.

^bMultivariate models control for all the gender, age, education, wealth, and urbanicity.

^cMultivariate models control for all the gender, education, wealth, and urbanicity. Data are β's (95% CI) for global cognitive function z scores. REF: reference category. Statistically significant associations at the 0.05 level are marked in bold.

covariates. The continuous CVH index was not associated with cognitive function in either the bivariate or multivariate models. Regarding the 0–4 categorical CVH index, in the bivariate linear regression, participants with the best levels of CVH (score 4) had cognitive function scores 0.47 SD higher than those with a score of 2 (p < 0.05). After controlling for covariates, participants with best CVH (score 4) had higher cognitive function scores than those with a CVH score of 0 (0.41 SD),

1 (0.39 SD), and 2 (0.56 SD; p < 0.05 for all differences). The interaction between CVH and age group was statistically significant (p < 0.001). The association between CVH and cognitive function was only present among participants 50–64 years old in which best CVH (score 4) was associated with higher cognitive function scores compared to CVH scores of 0 (0.56 SD), 1 (0.49 SD), 2 (0.68 SD), and 3 (0.42 SD; p < 0.05 for all differences).

Table 4. Relationship of individual CVH factors with global cognitive function

| | BIVARIATE LINEAR REGRESSION ^a | MULTIVARIATE LINEAR REGRESSION ^b |
|-------------------------------|---|--|
| Tobacco use (Ideal REF) | | |
| Poor/intermediate | 0.33 (0.14; 0.52) | 0.17 (0.01;0.33) |
| Physical activity (Ideal REF) | | |
| Poor/intermediate | -0.17 (-0.33;-0.01) | -0.04 (-0.168;0.08) |
| Body mass index (Ideal REF) | | |
| Poor/intermediate | 0.14 (-0.08;0.36) | -0.01 (-0.19;0.19) |
| Blood pressure (Ideal REF) | | |
| Poor/intermediate | -0.29 (-0.53;-0.04) | -0.27 (-0.49;-0.05) |

^aBivariate associations between each of the variables in the column and global cognitive function independently.

^bMultivariate models control for all the gender, age, education, wealth, and urbanicity. Data are β 's (95% CI) for global cognitive functionz scores. REF: reference category. Statistically significant associations at the 0.05 level are marked in bold.

Table 4 shows the bivariate and multivariate associations of global cognitive function with the individual CVH factors (smoking, exercise, BMI, and blood pressure). Poor/intermediate levels of smoking were associated with 0.33 *SD* higher of cognitive function compared to ideal levels in bivariate and 0.17 *SD* in multivariate associations ($p < 0.05$ each). In bivariate associations, ideal levels of exercise were associated with 0.17 *SD* higher of cognitive function than poor/intermediate levels, but the association disappeared after controlling for covariates. BMI was not associated with cognitive function in either model. Ideal blood pressure levels were associated with 0.29 *SD* higher of cognitive function compared to poor/intermediate levels in the bivariate model and 0.27 *SD* in the multivariate model.

Discussion

This study has examined the cross-sectional association between an index of ideal levels of modifiable CVH factors and cognitive function among a representative sample of Mexican older adults. Findings suggest that CVH is positively but non-linearly associated with cognitive function in this population. In particular, participants with the best levels of CVH have higher cognitive function than those at lower levels. These associations are modified by age and are only present among people aged 50–64 years but not older.

The present study adds to the growing evidence that CVH is important for cognitive function (Reis *et al.*, 2013; Crichton *et al.*, 2014; Thacker *et al.*, 2014; Gardener *et al.*, 2016; González *et al.*, 2016). The exclusion of participants with stroke suggests that there might be alternative vascular mechanisms for cognitive impairment other than

stroke (Kulshreshtha *et al.*, 2013). Contrary to findings from some previous studies using the AHA definition of CVH, we found that only the best levels of CVH were associated with substantially higher cognitive function. This data contrasts with linear cross-sectional associations found among Latinos aged 45–74 years, and longitudinal associations in the general population aged 18–30 years in the USA (Reis *et al.*, 2013; González *et al.*, 2016). The results also contrast with a longitudinal study among Americans 45 years and older that found that cognitive impairment was highest for those with poor CVH but the same for those with intermediate and high levels of CVH (Thacker *et al.*, 2014). In our study, however, CVH was measured using only four out of the seven AHA components of CVH, making comparisons with other studies different. We also found that the association between the categorical index of CVH and cognitive function was stronger than associations with individual CVH components. This finding supports the idea that dementia prevention trials should focus on multiple cardiovascular risk reduction (Olanrewaju *et al.*, 2015). These results are also in line with the AHA notion that prevention should not merely focus on preventing poor CVH levels but also promote optimal levels (Lloyd-Jones *et al.*, 2010).

These results are the first attempt to examine the association between an AHA-like index of ideal levels of modifiable CVH factors and cognitive function in the Mexican population. The importance of using an AHA-like approach of CVH relies upon including biomarkers and behaviors that are modifiable and account for optimal levels of CVH (Lloyd-Jones *et al.*, 2010). Therefore, the index used in the present study has more direct implications for health promotion and disease prevention than studies assessing the

association with single factors, (Biessels *et al.*, 2006; Cataldo *et al.*, 2010; Bherer *et al.*, 2013) examining solely poor levels of CVH (Biessels *et al.*, 2006; Reitz *et al.*, 2007) or including non-modifiable components (Unverzagt *et al.*, 2011).

This study shows that the association between CVH and cognitive function is only present among participants 50 – 64 years old. These findings are consistent with the literature as for example, BMI in midlife has been shown to be associated with dementia and cognitive function in later life, whereas BMI measured later in life has an inverse association with cognitive impairment (Fitzpatrick *et al.*, 2009). Similarly, midlife hypertension has been shown to be an important modifiable risk factor for late life cognitive decline and dementia (Whitmer *et al.*, 2005). However, the association in older ages remains unclear (Verghese *et al.*, 2003; Kloppenborg *et al.*, 2008). This study therefore adds to the evidence on the importance of timing and supports the idea that midlife may be a critical period for conducting CVH interventions to reduce dementia risk (Kloppenborg *et al.*, 2008; Gorelick *et al.*, 2011).

Findings from this study may also apply to many older adults living in the USA. In fact, a higher AHA CVH index score was cross-sectionally associated with better cognitive function among USA Latinos of whom 33.4% were of Mexican descent (González *et al.*, 2016). Currently, 34% of the 33.7 million Mexican Americans are Mexico-born (Gonzalez-Barrera and Lopez, 2013). Mexican Americans might share cultural and genetic characteristics related to CVH with the Mexican population, especially first generation ones. However, studies also suggest that the adaptation of Mexican Americans to the USA dominant culture might put them at a higher risk of dementia, as their risk of obesity, diabetes, sedentary lifestyle and smoking increases with the number of years lived in the USA (Goel *et al.*, 2004; Caballero, 2005; Kondo *et al.*, 2016). Generalizability of these findings to Mexican Americans may be threatened due to the Hispanic paradox in which there is a positive selection of immigrants from Mexico to the USA from the general population (Markides and Eschbach, 2005).

There are limitations to this study. First, the study lacked data on nutrition, glucose levels and cholesterol and therefore could not replicate the AHA definition of CVH (Lloyd-Jones *et al.*, 2010). Differences found between this and other studies using the AHA definition of CVH may be related to the incomplete composite score. Second, the CVH index has not been previously validated in the Mexican population. Third, the assessment

of CVH gives the same weight to the different domains, which might not represent their real contribution. Fourth, this study is cross-sectional, and therefore, causality cannot be inferred from the associations. In fact, associations such as the one between smoking and higher cognitive function have been shown to be an artifact of cross-sectional data, whereas longitudinal data shows associations in the opposite direction (Cataldo *et al.*, 2010). Fifth, urbanicity was predefined according to the way the areas were legally proclaimed to be, which ignores the participants' urban or rural migrations. However, 90.6% of the sample had lived in the same locality either always or most of their adult lives adding little information to the predefined urbanicity variable. Finally, the lower response rate (51%) may increase the risk of selection bias. Regarding potential public health implications, this study highlights the importance of optimal levels of CVH, especially in midlife as a potential means to improve brain health among Mexican older adults. If results are confirmed with longitudinal data, this will mean that greater effort needs to be made to prevent cognitive decline by promoting optimal levels of CVH as a whole as the current prevalence of best CVH levels is 2.2%. Comprehensive worksite wellness programs targeting weight, physical activity, blood pressure, and tobacco use might be optimal given that most Mexicans 50–64 years old are working and spend a significant part of their day at work. Workplace interventions have the potential to have economic and productivity benefits to employers in addition to health improvements (Baicker *et al.*, 2010). These interventions will work best if paired with improvements in other evidence-based population CVH strategies, including media and educational campaigns, labeling and consumer information, taxation, subsidies, and other economic incentives, local environmental changes, direct restrictions, and mandates (Mozaffarian *et al.*, 2012).

Conclusion

These findings add to the growing evidence that CVH is an important factor for cognitive health and is the first study in Mexico to address this association using an index of ideal levels of modifiable CVH factors. We found that the best levels of CVH were associated with higher cognitive function compared to other levels among stroke-free Mexican older adults. These results suggest that dementia-related policies in Mexico need to focus on optimizing CVH as a whole rather than simply preventing poor levels of isolated CVH factors. These findings also suggest that

improving CVH at midlife might be more beneficial for cognitive function than later in life. Future research is needed as CVH might be a key to slow down the devastating health and financial dementia consequences affecting individuals, families, and societies in Mexico.

Conflict of interest

None.

Description of authors' roles

J. Perales was involved in the conception and design of the work and carried out the analysis. J. Perales, L. Hinton, J. Burns, and E. Vidoni were involved in the interpretation of data. The first version of the manuscript was written by J. Perales and was subsequently improved by L. Hinton, J. Burns, and E. Vidoni with important intellectual contributions. All authors have approved the final version.

Acknowledgments

JP is thankful to the SAGE teams in Mexico and the World Health Organization. He is also grateful for the chance to attend the Mexico capacity building sessions granted by his former team at the Parc Sanitari Sant Joan de Déu, Barcelona and funded by the European Commission and the Instituto Carlos III through the COURAGE in Europe project. This study was supported by the World Health Organization and the US National Institute on Aging through Interagency Agreements (OGHA 04034785; YA1323-08-CN-0020; Y1-AG-1005-01) and through a research grant (R01-AG034479). Research reported in this publication was also supported by the National Institute on Aging of the National Institutes of Health under Award Number P30AG035982. The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health.

References

- Al Hazzouri, A. Z., Haan, M. N., Neuhaus, J. M., Pletcher, M., Peralta, C. A. and López, L.** (2013). Cardiovascular risk score, cognitive decline, and dementia in older Mexican Americans: the role of sex and education. *Journal of the American Heart Association*, 2, e004978.
- Baicker, K., Cutler, D. and Song, Z.** (2010). Workplace wellness programs can generate savings. *Health Affairs*, 29, 304–311.
- Bherer, L., Erickson, K. I. and Liu-Ambrose, T.** (2013). A review of the effects of physical activity and exercise on cognitive and brain functions in older adults. *Journal of Aging Research*, 2013. doi:10.1155/2013/657508.
- Biessels, G. J., Staekenborg, S., Brunner, E., Brayne, C. and Scheltens, P.** (2006). Risk of dementia in diabetes mellitus: a systematic review. *The Lancet Neurology*, 5, 64–74.
- Bonvecchio, A., Fernández-Gaxiola, A., Plazas, M., Kaufer-Horwitz, M., Pérez, A. and Rivera, J.** (2015). Nutrition and physical activity guidelines for overweight and obesity in the Mexican population [Guías alimentarias y de actividad física en contexto de sobrepeso y obesidad en la población mexicana]. Documento de Postura. Academia Nacional de Medicina (ANM). Intersistemas, México.
- Bull, F. C., Maslin, T. S. and Armstrong, T.** (2009). Global physical activity questionnaire (GPAQ): nine country reliability and validity study. *Journal of Physical Activity and Health*, 6, 790–804.
- Caballero, A. E.** (2005). Diabetes in the Hispanic or Latino population: genes, environment, culture, and more. *Current Diabetes Reports*, 5, 217–225.
- Cataldo, J. K., Prochaska, J. J. and Glantz, S. A.** (2010). Cigarette smoking is a risk factor for Alzheimer's disease: an analysis controlling for tobacco industry affiliation. *Journal of Alzheimer's Disease*, 19, 465–480.
- Crichton, G. E., Elias, M. F., Davey, A. and Alkerwi, A. A.** (2014). Cardiovascular health and cognitive function: the Maine-Syracuse longitudinal study. *PLoS One*, 9, e89317.
- Dong, C., Rundek, T., Wright, C. B., Anwar, Z., Elkind, M. S. and Sacco, R. L.** (2012). Ideal cardiovascular health predicts lower risks of myocardial infarction, stroke, and vascular death across whites, blacks and Hispanics: the northern Manhattan study. *Circulation*, 125, 2975–2984. doi:10.1161/CIRCULATIONAHA.111.081083.
- Escobedo, P. S. and Hollingworth, L.** (2009). Annotations on the use of the Mexican norms for the WAIS-III. *Applied Neuropsychology*, 16, 223–227.
- Fitzpatrick, A. L. et al.** (2009). Midlife and late-life obesity and the risk of dementia: cardiovascular health study. *Archives of Neurology*, 66, 336–342.
- Gardener, H. et al.** (2016). Ideal cardiovascular health and cognitive aging in the Northern Manhattan study. *Journal of the American Heart Association*, 5, e002731.
- Goel, M. S., Mccarthy, E. P., Phillips, R. S. and Wee, C. C.** (2004). Obesity among US immigrant subgroups by duration of residence. *JAMA*, 292, 2860–2867.
- Gonzalez-Barrera, A. and Lopez, M. H.** (2013). *A Demographic Portrait of Mexican-Origin Hispanics in the United States*. Washington, DC: Pew Hispanic Center.
- González, H. M. et al.** (2016). Life's simple 7's cardiovascular health metrics are associated with Hispanic/Latino neurocognitive function: HCHS/SOL results. *Journal of Alzheimer's Disease*, 53, 955–965.
- Gorelick, P. B. et al.** (2011). Vascular contributions to cognitive impairment and dementia. *Stroke*, 42, 2672–2713.
- Hoos, T., Espinoza, N., Marshall, S. and Arredondo, E. M.** (2012). Validity of the global physical activity

- questionnaire (GPAQ) in adult Latinas. *Journal of Physical Activity and Health*, 9, 698–705.
- IBM Corp. Released** (2013). *IBM SPSS Statistics for Windows. Version 22.0*. Armonk, NY: IBM Corp.
- Instituto Nacional De Salud Pública (INSP)** (2014). *Study on Global Ageing and Adult Health (SAGE) Wave 1: Mexico National Report*. London, UK: Instituto Nacional De Salud Pública (INSP).
- Kloppenborg, R. P., Van Den Berg, E., Kappelle, L. J. and Biessels, G. J.** (2008). Diabetes and other vascular risk factors for dementia: which factor matters most? A systematic review. *European Journal of Pharmacology*, 585, 97–108.
- Kondo, K. K., Rossi, J. S., Schwartz, S. J., Zamboanga, B. L. and Scaf, C. D.** (2016). Acculturation and cigarette smoking in Hispanic women: a meta-analysis. *Journal of Ethnicity in Substance Abuse*, 15, 46–72.
- Kowal, P. et al.** (2012). Data resource profile: the World Health Organization Study on Global Ageing and Adult Health (SAGE). *International Journal of Epidemiology*, 41, 1639–1649.
- Kulshreshtha, A. et al.** (2013). Life's simple 7 and risk of incident stroke. *Stroke*, 44, 1909–1914.
- Lloyd-Jones, D. M. et al.** (2010). Defining and setting national goals for cardiovascular health promotion and disease reduction. *Circulation*, 121, 586–613.
- Markides, K. S. and Eschbach, K.** (2005). Aging, migration, and mortality: current status of research on the Hispanic paradox. *The Journals of Gerontology: Series B: Psychological Sciences and Social Sciences*, 60, S68–S75.
- Mejia-Arango, S. and Gutierrez, L. M.** (2011). Prevalence and incidence rates of dementia and cognitive impairment no dementia in the Mexican population: data from the Mexican Health and Aging Study. *Journal of Aging and Health*, 23, 1050–1074.
- Mejía-Arango, S., Miguel-Jaimes, A., Villa, A., Ruiz-Arregui, L. and Gutiérrez-Robledo, L. M.** (2007). Cognitive impairment and associated factors in older adults in Mexico. *Salud Pública de México*, 49, S475–S481.
- Morris, J. C., Mohs, R., Rogers, H., Fillenbaum, G. and Heyman, A.** (1988). Consortium to establish a registry for Alzheimer's disease (CERAD) clinical and neuropsychological assessment of Alzheimer's disease. *Psychopharmacology Bulletin*, 24, 641–652.
- Mozaffarian, D. et al.** (2012). Population approaches to improve diet, physical activity, and smoking habits: a scientific statement from the American Heart Association. *Circulation*, 126, 1514–1563. doi:10.1161/CIR.0b013e318260a20b.
- Olanrewaju, O., Clare, L., Barnes, L. and Brayne, C.** (2015). A multimodal approach to dementia prevention: a report from the Cambridge Institute of Public Health. *Alzheimer's & Dementia: Translational Research & Clinical Interventions*, 1, 151–156.
- Ordúñez, P., Silva, L. C., Rodríguez, M. P. and Robles, S.** (2001). Prevalence estimates for hypertension in Latin America and the Caribbean: are they useful for surveillance? *Revista Panamericana de Salud Pública*, 10, 226–231.
- Ostrosky-Solís, F., Ardila, A. and Rosselli, M.** (1999). NEUROPSI: a brief neuropsychological test battery in Spanish with norms by age and educational level. *Journal of the International Neuropsychological Society*, 5, 413–433.
- Pearson, T. A. et al.** (2013). American heart association guide for improving cardiovascular health at the community level, 2013 update. *Circulation*, 127, 1730–1753. doi:10.1161/CIR.0b013e31828f8a94.
- Prince, M., Bryce, R., Albanese, E., Wimo, A., Ribeiro, W. and Ferri, C. P.** (2013). The global prevalence of dementia: a systematic review and metaanalysis. *Alzheimer's & Dementia*, 9, 63–75. e2.
- Reis, J. P. et al.** (2013). Cardiovascular health through young adulthood and cognitive functioning in midlife. *Annals of Neurology*, 73, 170–179.
- Reitz, C., Tang, M.-X., Manly, J., Mayeux, R. and Luchsinger, J. A.** (2007). Hypertension and the risk of mild cognitive impairment. *Archives of Neurology*, 64, 1734–1740.
- Secretaría de Salud** (1999). *Official Mexican Guidelines for the Prevention, Treatment and Control of Hypertension [Norma Oficial Mexicana, NOM-030-SSA2-1999, Para la Prevención, Tratamiento y Control de la Hipertensión Arterial]*. Mexico City, Mexico: Secretaría de Salud.
- Secretaría de Salud** (2009). *Official Mexican Guidelines for the Prevention, Treatment and Control of Addictions [Norma Oficial Mexicana, NOM-028-SSA2-2009, Para la Prevención, Tratamiento y Control de las Adicciones]*. Mexico City, Mexico: Secretaría de Salud.
- Shaw, J. E., Sicree, R. A. and Zimmet, P. Z.** (2010). Global estimates of the prevalence of diabetes for 2010 and 2030. *Diabetes Research and Clinical Practice*, 87, 4–14.
- Silvia, M.-A. and Clemente, Z.-G.** (2011). Diabetes mellitus as a risk factor for dementia in the Mexican elder population. *Revista de Neurologia*, 53, 397.
- Sosa, A. L. et al.** (2009). Population normative data for the 10/66 Dementia Research Group cognitive test battery from Latin America, India and China: a cross-sectional survey. *BMC Neurology*, 9, 48.
- Thacker, E. L. et al.** (2014). The American Heart Association life's simple 7 and incident cognitive impairment: the REasons for Geographic And Racial Differences in Stroke (REGARDS) study. *Journal of the American Heart Association*, 3, e000635.
- Tulsky, D. and Zhu, J.** (2003). *Escala Wechsler de Inteligencia para Adultos-III*. Mexico, DF: El Manual Moderno.
- Unverzagt, F. et al.** (2011). Vascular risk factors and cognitive impairment in a stroke-free cohort. *Neurology*, 77, 1729–1736.
- Üstun, T., Chatterji, S., Mechbal, A. and Murray, C.** (2005). Quality assurance in surveys: standards, guidelines and procedures. In *Household Sample Surveys in Developing and Transition Countries* (pp. 199–230). New York, NY: United Nations.
- Verghese, J., Lipton, R., Hall, C., Kuslansky, G. and Katz, M.** (2003). Low blood pressure and the risk of dementia in very old individuals. *Neurology*, 61, 1667–1672.

- Wechsler, D.** (2001). *Escala Wechsler de Inteligencia Para Adultos-III (WAIS-III)*. Mexico, DF: El Manual Moderno.
- Wechsler, D., Coalson, D. L. and Raiford, S. E.** (1997). *WAIS-III: Wechsler Adult Intelligence Scale*. San Antonio, TX: Psychological Corporation.
- Whitmer, R. A., Sidney, S., Selby, J., Johnston, S. C. and Yaffe, K.** (2005). Midlife cardiovascular risk factors and risk of dementia in late life. *Neurology*, 64, 277–281.
- World Health Organization** (2012). *Dementia: A Public Health Priority*. Geneva, Switzerland: World Health Organization.
- World Health Organization** (2015). *Country Statistics and Global Health Estimates*. Mexico: World Health Organization.
- World Obesity Federation** (2017). *World Obesity: Obesity Prevalence Worldwide-Adults*. Available at: <http://www.worldobesity.org/data/map/overview-adults#country=CAN>; last accessed 6 December 2017.
- Yaffe, K., Haan, M., Blackwell, T., Cherkasova, E., Whitmer, R. A. and West, N.** (2007). Metabolic syndrome and cognitive decline in elderly Latinos: findings from the Sacramento Area Latino study of aging study. *Journal of the American Geriatrics Society*, 55, 758–762.

Convergent and concurrent validity of a report- versus performance-based evaluation of everyday functioning in the diagnosis of cognitive disorders in a geriatric population

Elise Cornelis,^{1,2,3} Ellen Gorus,^{1,2,4} Karen Van Weverbergh,⁴ Ingo Beyer^{1,2,4}
and Patricia De Vriendt^{2,3,4}

¹Department of Geriatrics, Universitair Ziekenhuis Brussel, Brussels, Brussels Capital, Belgium

²Frailty in Ageing Research Group (FRIA), Vrije Universiteit Brussel, Brussels, Brussels Capital, Belgium

³Department of Occupational Therapy, Artevelde University College Ghent, Ghent, East Flanders, Belgium

⁴Department of Gerontology (GERO), Vrije Universiteit Brussel, Brussels, Brussels Capital, Belgium

ABSTRACT

Background: Several methods have been developed to evaluate activities of daily living (ADLs) in mild cognitive impairment (MCI) and mild dementia. This study evaluated the convergent and concurrent validity between (1) two report-based methods (the advanced (a)- and instrumental (i)-ADL tools) and (2) a performance-based method (the Naturalistic Action Test (NAT)) to check if their ability to differentiate between cognitively healthy comparisons (HCs), persons with MCI, and persons with mild Alzheimer's disease (AD) are comparable to each other.

Method: This was a cross-sectional study, undertaken in a geriatric day hospital. The participants comprised community-dwelling HCs ($n = 21$, median age 78.0 years, 61.9% female), MCI ($n = 20$, median age 79.5 years, 55.0% female), and AD ($n = 20$, median age 80.0 years, 85.0% female) adults. A diagnostic procedure for neurocognitive disorders was employed. In addition, the a- and i-ADL tools and the NAT were administered separately by blinded raters.

Results: The NAT and both the a- and i-ADL tools showed significant differences between HCs, MCI, and AD participants. Convergent validity showed moderate to strong significant correlations between the NAT, and a- and i-ADL tools (range -0.583 to -0.663 ; $p < 0.01$). Concurrent validity showed that the NAT (AUC 0.809–1.000) and the a- and i-ADL tools (AUC 0.739–0.964) presented comparable discriminatory accuracy ($p = 0.0588$).

Conclusions: In contrast to prior studies comparing report-based and performance-based methods of assessing ADL, this study indicates that the NAT and the a- and i-ADL tools have strong convergent and concurrent validity, and appear to have similar discriminatory power in differentiating between HCs, MCI, and AD.

Key words: mild cognitive impairment (MCI), mild dementia, activities of daily living (ADLs), assessment

Introduction

The evaluation of functional decline in activities of daily living (ADLs) is pivotal in diagnosing cognitive disorders in older persons. A loss of autonomy in ADLs is a core feature of dementia and is required to establish a diagnosis (McKhann *et al.*, 2011). Moreover, it is recognized that

mild changes in advanced (a-)ADLs, which covers activities that go beyond independent living and express a personal engagement, such as leisure, self-development activities, or (semi-) professional work (Reuben *et al.*, 1990), and instrumental (i-)ADLs, including activities essential to maintain independent living, such as cooking, shopping, and managing medication, are already experienced in the earlier stages of the disease, as in mild cognitive impairment (MCI) (Gold, 2012). MCI is conceptualized as a transitional period between healthy cognitive aging and mild dementia (Petersen, 2004; Petersen *et al.*, 2014) and its identification is helpful for a timely diagnosis, offering opportunities for early intervention (Dubois *et al.*, 2016). Assessing

Correspondence should be addressed to: Prof. Patricia De Vriendt, Frailty in Ageing Research Group (FRIA) and Department of Gerontology (GERO), Faculty of Medicine and Pharmacy, Vrije Universiteit Brussel, Laarbeeklaan 103, B-1090 Brussels, Belgium. Phone: +32-479-65-41-10; Fax: +32-447-63-64. Email: patricia.de.vriendt@vub.ac.be. Received 25 Oct 2017; revision requested 19 Nov 2017; revised version received 22 Feb 2018; accepted 22 Feb 2018. First published online 22 March 2018.

ADLs is not only useful in diagnostics but recent evidence also states that disabilities – especially in a- and i-ADLs – are strong predictors of future dementia (Fauth *et al.*, 2013), and contribute to a higher and faster risk for conversion from MCI to mild dementia (Anstey *et al.*, 2013).

Many attempts have been made to find sensitive but easy-to-administer and inexpensive assessments to identify functional decline, and hence to allow the differentiation between MCI and mild dementia (Gold, 2012; Marshall *et al.*, 2012; Jekel *et al.*, 2015). In clinical practice, everyday functioning is most commonly assessed through interviews with patients or proxies.

Report-based scales, such as the Lawton Scale (Lawton and Brody, 1969) and many other instruments, are usually quick and easy to administer (Jekel *et al.*, 2015; Wesson *et al.*, 2016). Self-report assessment in which a patient is asked to report on everyday functioning is the most direct and simple method for gathering information (Sikkes and Rotrou, 2014). However, for individuals with cognitive disorders, an informant-report seems to be superior as patients may be too impaired to report accurately on their ADLs and may over- or underestimate functional limitations (Marshall *et al.*, 2012; Puente *et al.*, 2014). Therefore, an informant-report is considered more reliable, providing a good representation of everyday performance (Marshall *et al.*, 2012; Schmitter-Edgecombe *et al.*, 2014). However, informant-report scales might also be prone to error, and reporter bias has been found due to the caregiver burden and distress (Puente *et al.*, 2014; Schmitter-Edgecombe *et al.*, 2014). Thus, most report-based tools have limitations for diagnostic purposes. Moreover, most tools have scoring systems with poor sensitivity in detecting mild functional impairments (Fieo *et al.*, 2011) and show a lack of psychometric value (Gold, 2012; Jekel *et al.*, 2015; Wesson *et al.*, 2016).

To address the concerns related to report-based scales, performance-based measures, such as the Assessment of Motor and Process Skills (AMPS) (Fisher, 1999), the Large Allen's Cognitive Levels Screen-5 (Allen *et al.*, 2007), and the Naturalistic Action Test (NAT) (Schwartz *et al.*, 2002), can be used. In performance-based evaluations, individuals are asked to perform everyday tasks while being observed by a trained rater. In this way, it is argued that performance-based scales provide a more objective measure of ADLs (Puente *et al.*, 2014; Schmitter-Edgecombe *et al.*, 2014; Jekel *et al.*, 2015; Wesson *et al.*, 2016). However, performance-based scales can also be influenced by the motivation and behavior of the patient, and they are time-consuming and

expensive to administer (Schmitter-Edgecombe *et al.*, 2014; Jekel *et al.*, 2015; Wesson *et al.*, 2016). Moreover, performance-based scales often only allow observation of a small excerpt of everyday functioning, can sometimes solely be administered by trained occupational therapists (e.g. the AMPS) (Fisher, 1999), and are mostly performed in artificial environments outside the daily routine of individuals (Gold, 2012). Some of them provide limited information on psychometric properties and have ceiling effects, and the differentiation between older persons with normal cognition, MCI, and mild dementia is not sufficiently studied (Wesson *et al.*, 2016).

The advantages and limitations of report-based and performance-based measures have led to discussions regarding the most suitable method for evaluating ADLs. Currently, there is no gold standard that is time efficient and easy to administer (Reppermund *et al.*, 2016). Only a limited number of studies have compared report-based and performance-based measures in the same sample of older persons and have found limited correlations between the two measures (Schmitter-Edgecombe *et al.*, 2014; Rycroft *et al.*, 2017). This results in significant differences between report-based and performance-based information on ADLs (Sager *et al.*, 1992; Bean *et al.*, 2011; Nielsen *et al.*, 2016; Roedersheimer *et al.*, 2016). Recently, Wesson *et al.* (2016) recommended the need for studies to refine the psychometric properties of existing instruments and Jekel *et al.* (2015) stressed the need for studies comparing report-based and performance-based evaluations in the same sample to allow a valid and reliable assessment of ADLs in MCI and mild dementia.

Therefore, in this study a report-based and performance-based evaluation, both with good psychometric values for diagnosing mild cognitive disorders, were compared within the same study sample. For the report-based evaluation, this study used the recently developed and validated advanced (a-)ADL tool (De Vriendt *et al.*, 2013; De Vriendt *et al.*, 2015) and the instrumental (i-)ADL tool (Cornelis *et al.*, 2017). For the performance-based evaluation, this study used the NAT, which evaluates a broad range of performance skills in the a- and i-ADLs frameworks. The NAT is feasible for administration in clinical settings (by any trained clinician and without the need for expensive materials), has been validated in dementia patients (Giovannetti *et al.*, 2002a), and has recently been used to discriminate MCI from mild dementia.

This study aimed to evaluate the convergent and concurrent validity of the a- and i-ADL tools and the NAT in order to compare their accuracy in making a diagnostic differentiation between

cognitively healthy older persons and those with MCI and mild dementia. Based on prior research findings, we might expect to find poor to moderate correlations between the report-based and the performance-based measurement tools, wherein, the latter having the best diagnostic accuracy. However, in our previous studies (De Vriendt *et al.*, 2013; De Vriendt *et al.*, 2015; Cornelis *et al.*, 2017) the a- and i-ADL tools present good to excellent ability in discriminating between HCs, MCI, and Alzheimer's disease (AD) patients; so, we hypothesized that both the a- and i-ADL tools and the NAT might have similar diagnostic power.

Methods

Participants and procedure

Three groups of community-dwelling older persons (≥ 65 years) were recruited consecutively through the geriatric day hospital of an academic teaching hospital (UZ Brussel, Belgium): patients with MCI, patients with mild to moderate AD, and cognitively healthy comparisons (HCs).

Patients with MCI and AD underwent a standardized multidisciplinary diagnostic procedure, including a physical and neurological examination, clinical history taking, functional evaluation of basic (b-) and i-ADLs, using the Katz index (Katz, 1963) and the Lawton scale (Lawton and Brody, 1969), respectively, neuropsychological assessment, extensive laboratory blood testing, and imaging of the brain by CT or MRI scan, considered good clinical practice (Hugo and Ganguli, 2014). Diagnosis was based on the clinical consensus of the multidisciplinary team. Patients with MCI fulfilled the diagnostic criteria for a-MCI (Petersen, 2004). Patients with AD fulfilled the criteria of the National Institute of Neurological and Communicative Disorders and Stroke and the AD and Related Disorders Association (NINCDS-ADRDA) (McKhann, 1984). When the presence of major depression was presumed, this ruled out the diagnosis of MCI or AD at that stage.

HCs represented geriatric patients who visited the geriatric day hospital for diagnosis or the treatment of conditions other than cognitive disorders (e.g. osteoporosis). They were recruited separately from the diagnostic process for MCI and AD, but were evaluated using the same neuropsychological assessment and functional evaluation as for MCI and AD.

For all participants, exclusion criteria were any acute pathology, sensory, or communicative impairments that precluded them from participating in the assessments, and a history of major psychiatric illness or any pathology of the central

nervous system other than MCI or AD (e.g. stroke, epilepsy). Patients with MCI or AD were not included if they were not accompanied by a reliable informant. Informants were considered reliable when they were spouses, family, or close friends who could provide accurate information about the patients' ADL. The informant's ability to provide accurate information was operationalized by asking each person with MCI or AD if his or her informant was someone who knew them well and could provide accurate information about their daily life. For HCs, additional exclusion criteria were a score $< 26/30$ on the Mini-Mental State Examination (MMSE) (Folstein *et al.*, 1975), a score $< 80/105$ on the Cambridge Cognitive Test (CAMCOG) (Gallagher *et al.*, 2010), or a score $< 18/27$ on the CAMCOG memory section and a self- or informant-based history of functional or cognitive deficits suggestive of MCI or AD.

After the participants completed the diagnostic procedure, a trained occupational therapist conducted the NAT, and another trained occupational therapist conducted the a- and i-ADL tools, in which self-report was used for HCs, and an informant-report was obtained for MCI and AD. The occupational therapists were blinded to the outcomes of the other evaluations and the diagnosis. Both the NAT and the a- and i-ADL tools were always administered by the same occupational therapist on the same day. For the HCs, the a- and i-ADL tools and the NAT were administered consecutively. For MCI and AD, the a- and i-ADL tools and NAT were conducted at the same time.

The naturalistic action test

The NAT comprises three tasks of increasing difficulty, including preparing toast and coffee, wrapping a present, and packing a lunch box and a schoolbag (see Table 1). Instructions, cues, termination procedures, and the placement of objects are standardized (Schwartz *et al.*, 2002). Participants are scored for accomplishing each task (i.e. the steps they perform). In addition, errors during performance are recorded and combined with the accomplishment score into a score ranging 0–6 for each task (higher scores signify better performance). The maximum total score is 18. Based on a study with a sample of persons with dementia, Sestito *et al.* (2005) suggested a score of ≤ 13 as being in the impaired range. Recently, Bruce *et al.* (2016) recommended a cut-off point of ≥ 11 to differentiate MCI from mild dementia (AUC of 0.808). The inter-rater reliability of the NAT is high, both for scoring accomplishment of tasks (median weighted kappa of 0.98) and

Table 1. Description of the content of the NAT and the a- and i-ADL tools

| TASKS AND INSTRUCTIONS OF THE NAT | |
|--|---|
| NAT task 1 toast and coffee | Make a toast with butter and jelly/jam, and instant coffee with cream/milk and sugar. |
| NAT task 2 gift | Wrap a gift as a present. |
| NAT task 3 lunchbox and school bag | Prepare and pack a lunchbox with sandwich, drink, and snack/biscuits and pack a school bag with ring binder, notebook, and stocked pencil case. Ring bell attached to the underside of table top after completing each task. |
| ACTIVITIES ASSESSED IN THE I-ADL TOOL | ACTIVITIES ASSESSED IN THE A-ADL TOOL |
| Using communication devices and techniques | Freezing or pickling vegetables |
| Using transportation | Baking bread, cakes |
| Shopping | Cooking complex meals |
| Preparing meals | Try out new dishes |
| Doing housework | Making jam |
| Washing and drying clothes | Using a dish washer |
| Caring for household objects | Using an oven |
| Maintaining one's health | Using a coffee machine |
| Basic economic transactions | Using a kitchen aid |
| | Using a washing machine |
| | Using a drying machine |
| | Using a radio/CD |
| | Using a TV |
| | Using video/DVD |
| | Using a camera |
| | Using a lawn mower |
| | Using an electric saw |
| | Using a high pressure cleaner |
| | Use of manuals explaining daily technology |
| | Puzzles and brainteasers |
| | PC programs |
| | Use of internet |
| | Use of agenda |
| | Reading books |
| | Reading professional or educational literature |
| | Writing books, poems, articles |
| | Crafts |
| | Playing music instruments |
| | Practicing arts |
| | Electronically banking |
| | Complex administration |
| | Using a cell phone |
| | Writing an e-mail or letter |
| | Sports |
| | Riding a bicycle |
| | Transportation by motorized vehicles |
| | Self-development/self-realization/self-educational activities |
| | To go on a holiday |
| | To help (in the business) of the children |
| | Take care of partner |
| | Take care of (great) grand children |
| | Take care of pets |
| | Caring for household objects |
| | Semi-professional work |
| | Organizing events |
| | To make and keep appointments |
| | To take part in meetings, conversations |

recording errors (median percentage agreement 95%) (Schwartz *et al.*, 2002).

The a- and i-ADL tools

The a-ADL tool (De Vriendt *et al.*, 2013; De Vriendt *et al.*, 2015) and i-ADL tool (Cornelis *et al.*, 2017), both comprise semi-structured interviews that use the terminology and scoring system of the International Classification of Functioning (ICF) (World Health Organization, 2001). The a-ADL tool evaluates complex activities, such as hobbies or using technologies, whereas the i-ADL tool evaluates household activities, such as preparing a meal and doing groceries (see Table 1). These two tools only take relevant activities into account, meaning activities currently or recently (the tools suggest by thinking back to the years since retirement) performed by the individual. If activities have not been carried out during the past years because they were not relevant for an individual, they are not taken into account. There is no cut-off for the total number of relevant activities. The relevant activities are evaluated on present difficulties, based on the narratives of the patient or proxy. The scoring system adapted the performance qualifiers of the ICF, consisting of a five-point scale ranging from 0 (no difficulty to perform) to 4 (complete difficulty or unable to perform). If a score of 1 or higher is assigned, the interviewer also determines the underlying causes of limited performance, such as physical (e.g., mobility problems) and cognitive problems (e.g., memory problems). Both tools result in a set of disability indices. A “global disability index” (DI) is calculated by taking into account all relevant activities related to all the activities that are limited and the severity of the limitation, determining a “cognitive disability index” (CDI), considering only activities limited due to cognitive problems, and a “physical disability index” (PDI), considering activities limited due to physical problems. All indices are expressed as percentages, with higher scores representing greater disability. The a- and i-ADL tools both show good to excellent psychometric properties and have the ability to discriminate between cognitively healthy older persons, MCI, and mild dementia (AUC in the range 0.791–0.968) (De Vriendt *et al.*, 2013; De Vriendt *et al.*, 2015; Cornelis *et al.*, 2017). De Vriendt *et al.* (2015) recommend a cut-off point of 47.9% for the a-ADL-DI (AUC of 0.802) and 27.4% for the a-ADL-CDI (AUC of 0.804) to differentiate MCI from mild dementia. Cornelis *et al.* (2017) recommend a cut-off point of 44.1% for the i-ADL-DI (AUC of 0.736) and 23.6% for the i-ADL-CDI (AUC of 0.805) to differentiate

MCI from mild dementia. The inter-rater reliability is excellent for the a-ADL-DI (ICC = 0.996, 95% CI (0.991–0.998)), a-ADL-CDI (ICC = 0.979, 95% CI (0.952–0.991)), i-ADL-DI (ICC = 0.986, 95% CI (0.968 ± 0.994)), and i-ADL-CDI (ICC = 0.986, 95% CI (0.969 ± 0.994)) (all $p < 0.001$) (De Vriendt *et al.*, 2013; Cornelis *et al.*, 2017).

Statistical analysis

Statistical analyses were performed in IBM SPSS for Mac (version 22.0) (SPSS Inc., Illinois, USA), with an α -level set two sided at $p < 0.05$ for all analyses. There were no missing data. Since most datasets were not normally distributed (Kolmogorov–Smirnov goodness-of-fit test $p < 0.05$), non-parametric tests were used. Data are reported as medians and interquartiles. The demographic and clinical characteristics, NAT scores, and indices of the a- and i-ADL tools are described. Differences between HCs, MCI, and AD were calculated using the Kruskal–Wallis test for continuous variables and the Chi-Squared test for categorical variables. Differences between HCs versus MCI, MCI versus AD, and HCs versus AD were tested with the Mann–Whitney U test. Convergent validity was assessed using correlation analyses performed with Spearman’s r between the indices of the a- and i-ADL tools and the NAT total score. Convergent validity was considered poor if the correlation coefficient was below 0.30, moderate if between 0.30 and 0.59, and good if greater than 0.60 (Andresen, 2000). Concurrent validity was evaluated by first calculating the receiver operating characteristic (ROC) curves and the AUC for the indices of the a- and i-ADL tools and the scores of the NAT to compare the ability of the instruments to differentiate between HCs, MCI, and AD. Finally, the ROC curves and the AUC for the indices of the a- and i-ADL tools and the NAT total score were compared using the method of DeLong *et al.* (1988) in MedCalc (version 14.8.1.0) (MedCalc Software, Mariakerke, Belgium).

Results

Participants’ characteristics

The demographic and clinical characteristics of the HCs ($n = 21$), MCI ($n = 20$), and AD ($n = 20$) groups are shown in Table 2. There were no significant differences between the groups in terms of age, gender, education, or the Katz index. As expected, the MMSE, CAMCOG, and memory score of the CAMCOG showed significantly higher scores among HCs than among

Table 2. Participants' characteristics

| | HC (<i>N</i> = 21) | MCI (<i>N</i> = 20) | AD (<i>N</i> = 20) | TEST STATISTICS ^a | POST-HOC TEST STATISTICS ^b | | |
|--|------------------------|-------------------------|------------------------|-----------------------------------|---|---|--|
| | | | | | HC VERSUS MCI | MCI VERSUS AD | HC VERSUS AD |
| Age Median (interquartile) | 78.0 (10.0) | 79.5 (4.0) | 80.0 (9.0) | $\chi^2(2) = 1.74$ p = 0.419 | | | |
| Gender Female (%) | 61.9 | 55.0 | 85.0 | $\chi^2(2) = 4.49$ p = 0.106 | | | |
| Education, years Median (interquartile) | 12.0 (4.0) | 12.0 (5.0) | 12.0 (3.0) | $\chi^2(14) = 17.07$ p = 0.252 | | | |
| MMSE (./30) Median (interquartile) | 29.0 (2.0) | 26.0 (5.0) | 22.5 (5.0) | $\chi^2(2) = 36.07$ p < 0.001 | <i>U</i> = 42.50, p < 0.001, <i>r</i> = 0.69 | <i>U</i> = 103.00, p = 0.008, <i>r</i> = 0.41 | <i>U</i> = 7.50, p < 0.001, <i>r</i> = 0.83 |
| CAMCOG (./105) Median (interquartile) | 94.0 (7.0) | 84.5 (7.0) | 75.0 (16.0) | $\chi^2(2) = 37.41$ p < 0.001 | <i>U</i> = 32.50, p < 0.001, <i>r</i> = 0.72 | <i>U</i> = 93.50, p = 0.003, <i>r</i> = 0.45 | <i>U</i> = 9.50, p < 0.001, <i>r</i> = 0.81 |
| CAMCOG memory (./27) Median (interquartile) | 22.0 (3.0) | 19.0 (4.5) | 13.0 (6.0) | $\chi^2(2) = 35.25$ p < 0.001 | <i>U</i> = 68.50, p < 0.001, <i>r</i> = 0.58 | <i>U</i> = 56.00, p < 0.001, <i>r</i> = 0.61 | <i>U</i> = 16.50, p < 0.001, <i>r</i> = 0.79 |
| b-ADL Katz-Index (./24) Median (interquartile) | 6.0 (0.0) | 6.0 (2.0) | 6.5 (2.0) | $\chi^2(2) = 5.62$ p = 0.060 | | | |
| i-ADL Lawton & Brody Scale (./27) Median (interquartile) | 25.0 (4.0) | 22.0 (7.0) | 17.5 (9.0) | $\chi^2(2) = 16.21$ p < 0.001 | <i>U</i> = 148.00, p = 0.102, <i>r</i> = 0.25 | <i>U</i> = 118.50, p = 0.026, <i>r</i> = 0.34 | <i>U</i> = 55.00, p < 0.001, <i>r</i> = 0.63 |

HC: Healthy comparison; MCI: mild cognitive impairment; AD: Alzheimer's disease; MMSE: Mini-Mental State Examination; CAMCOG: Cambridge examination for mental disorders of the elderly, cognitive part.

b-ADL according to the Katz-Index has a minimum score of 6/24; lower scores are indicating higher autonomy; i-ADL according to the Lawton & Brody Scale has a minimum score of 9/27; lower scores are indicating lower autonomy.

^aDifferences between groups tested with Kruskal–Wallis test for continuous variables and χ^2 for categorical variables.

^bGroup by group tested with Mann–Whitney *U* test.

MCI and AD participants, and MCI participants compared to AD participants. The Lawton scale showed significantly higher scores for HCs and MCI participants compared to AD participants.

NAT scores

Performances on the NAT were significantly poorer for those with AD compared to MCI, and for those with MCI compared to the HCs group (see Table 3). Tasks 1 and 3 of the NAT differed significantly between the groups: persons with AD performed worse than those with MCI, and those with MCI performed worse than the HCs. Task 2 differed significantly between the HCs and those with MCI, and between the HCs group and those with AD, but there was no significant difference in performance between MCI and AD participants.

Indices of the a- and i-ADL tools

The median performed a-ADL was 27 activities (25th percentile = 23 activities; 75th percentile = 30, 5 activities). The median performed i-ADL

was 9 activities (25th percentile = 8 activities; 75th percentile = 9 activities). The indices for the a-ADL tool were derived from at least 10 activities (which was the case for 2 participants). The indices for the i-ADL tool were derived from at least 6 activities (which was the case for 4 participants). All other participants reported more activities. As illustrated in Table 3, the DI and CDI of both a- and i-ADL showed significantly more severe deficits in the AD group than MCI patients, and in the MCI patients than the HCs group. The PDI of both a- and i-ADL showed no significant differences between the groups.

Convergent validity between the a- and i-ADL tools and the NAT

The total scores of the NAT showed a significant and strong correlation with the a-ADL-DI, a-ADL-CDI, and i-ADL-CDI (*r* ranging from -0.634 to -0.663 ; all *p* < 0.01), and a significant moderate correlation with the i-ADL-DI (*r* = -0.583 ; *p* < 0.01). The PDI of a- and i-ADL did not correlate

Table 3. NAT scores and indices of the a- and i-ADL tools

| | HC (N = 21) | MCI (N = 20) | AD (N = 20) | TEST STATISTICS ^a | POST-HOC TEST STATISTICS ^b | | |
|---|----------------|-----------------|-----------------|----------------------------------|---------------------------------------|--------------------------------------|-------------------------------------|
| | | | | | HC < MCI | MCI < AD | HC < AD |
| NAT scores | | | | | | | |
| NAT total (/18) Median (interquartile) | 16.0 (3.0) | 11.5 (4.0) | 8.0 (8.0) | $\chi^2(2) = 43.73$ p < 0.001 | U = 14.0, p < 0.001, r = 0.81 | U = 76.5, p = 0.001, r = 0.53 | U = 0.0, p < 0.001, r = 0.81 |
| NAT task 1 (/6) Median (interquartile) | 6.0 (0.0) | 4.5 (4.0) | 2.0 (4.0) | $\chi^2(2) = 31.11$ p < 0.001 | U = 96.0 p < 0.001, r = 0.56 | U = 99.0, p = 0.005, r = 0.44 | U = 20.0, p < 0.001, r = 0.83 |
| NAT task 2 (/6) Median (interquartile) | 6.0 (2.0) | 4.0 (2.0) | 4.0 (4.0) | $\chi^2(2) = 22.37$ p < 0.001 | U = 68.0 p < 0.001, r = 0.60 | U = 155.0, p = 0.212, r = 0.19 | U = 54.5, p < 0.001, r = 0.66 |
| NAT task 3 (/6) Median (interquartile) | 4.0 (2.0) | 3.0 (2.0) | 0.0 (2.0) | $\chi^2(2) = 35.81$ p < 0.001 | U = 53.5 p < 0.001, r = 0.67 | U = 75.0, p < 0.001, r = 0.54 | U = 19.0, p < 0.001, r = 0.80 |
| Indices of the a- and i-ADL tools | | | | | | | |
| a-ADL-DI, % Median (interquartile) | 15.4% (8.6) | 35.9% (35.1) | 58.1% (36.4) | $\chi^2(2) = 28.71$ p < 0.001 | U = 75.5 p < 0.001, r = 0.54 | U = 104.5, p = 0.009, r = 0.41 | U = 22.0, p < 0.001, r = 0.76 |
| a-ADL-CDI, % Median (interquartile) | 6.6% (13.7) | 22.9% (29.1) | 56.1% (33.0) | $\chi^2(2) = 31.26$ p < 0.001 | U = 90.5 p = 0.002, r = 0.48 | U = 74.5, p < 0.001, r = 0.53 | U = 15.0, p < 0.001, r = 0.79 |
| a-ADL-PDI, % Median (interquartile) | 2.7% (5.5) | 3.0% (11.0) | 5.9% (10.7) | $\chi^2(2) = 4.35$ p = 0.113 | | | |
| i-ADL-DI, % Median (interquartile) | 5.5% (14.5) | 23.4% (35.9) | 52.7% (43.3) | $\chi^2(2) = 22.99$ p < 0.001 | U = 99.0 p = 0.004, r = 0.45 | U = 118.0, p = 0.026, r = 0.35 | U = 35.5, p < 0.001, r = 0.71 |
| i-ADL-CDI, % Median (interquartile) | 0.0% (0.0) | 5.5% (17.5) | 42.6% (43.8) | $\chi^2(2) = 36.93$ p < 0.001 | U = 84.0 p < 0.001, r = 0.63 | U = 77.0, p = 0.001, r = 0.53 | U = 10.5, p < 0.001, r = 0.88 |
| i-ADL-PDI, % Median (interquartile) | 0.0% (10.2) | 1.3% (10.4) | 6.3% (19.4) | $\chi^2(2) = 4.27$ p = 0.118 | | | |

HC: Healthy Comparison; MCI: Mild Cognitive Impairment; AD: Alzheimer's disease; NAT: Naturalistic Action Test; a-ADL-DI: advanced activities of daily living-disability index; a-ADL-CDI: advanced activities of daily living-cognitive disability index; a-ADL-PDI: advanced activities of daily living-physical disability index; i-ADL-DI: instrumental activities of daily living-disability index; i-ADL-CDI: instrumental activities of daily living-cognitive disability index; i-ADL-PDI: instrumental activities of daily living-physical disability index.

^aDifferences between groups tested with Kruskal–Wallis test.

^bGroup by group tested with Mann–Whitney U test.

significantly with the total score of the NAT (all p > 0.05).

Concurrent validity between the a- and i-ADL tools and the NAT

Table 4 presents the results of the ROC curves for the NAT, and the indices of the a- and i-ADL tool. The AUC of the NAT total score ranges from 0.809 to 1.000 but has no significantly better accuracy than the DI and CDI of both a- and i-ADL (AUC ranges from 0.739 to 0.964) for differentiating between the three groups. The PDI of a- and i-ADL did not show significantly better accuracy than the NAT.

Discussion

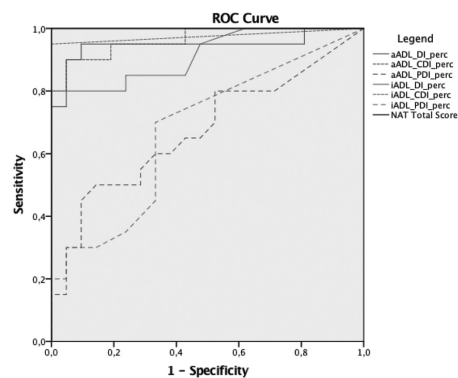
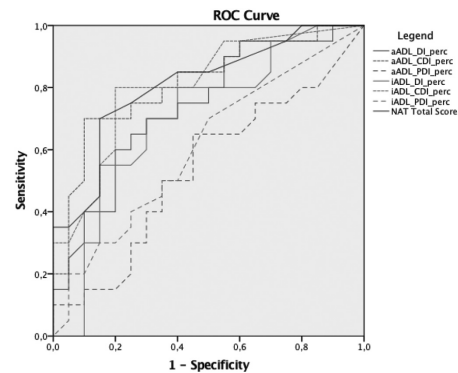
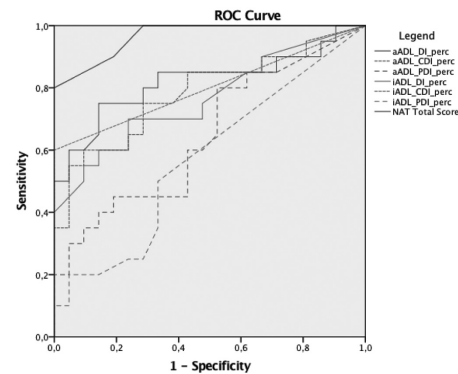
Report-based and performance-based measures have been developed to evaluate ADL in underpinning the diagnosis of cognitive disorders and to distinguish MCI from the earliest manifestations of dementia. Nevertheless, until now there has been no consensus on the most accurate measurement method. Studies comparing report-based and performance-based methods remain scarce (Jekel *et al.*, 2015). In this study we examined the convergent and concurrent validity and diagnostic accuracy between the NAT – as a performance-based method – and the a- and i-ADL tools – as report-based methods – in a sample of HCs, persons with MCI, and persons with mild dementia.

Table 4. Concurrent validity between the NAT and the a- and i-ADL tools

| HC VERSUS MCI | | | |
|-----------------|--------------|-------------|----------------------|
| | AUC (S.E.) | 95% CI | P-VALUE ^a |
| NAT total score | 0.967 (0.02) | 0.922–1.000 | |
| NAT task 1 | 0.771 (0.07) | 0.620–0.923 | |
| NAT task 2 | 0.838 (0.06) | 0.717–0.960 | |
| NAT task 3 | 0.873 (0.05) | 0.761–0.984 | |
| a-ADL-DI | 0.820 (0.07) | 0.682–0.959 | 0.210 |
| a-ADL-CDI | 0.785 (0.07) | 0.641–0.929 | 0.163 |
| a-ADL-PDI | 0.646 (0.08) | 0.476–0.817 | <0.001 |
| i-ADL-DI | 0.764 (0.07) | 0.615–0.913 | 0.058 |
| i-ADL-CDI | 0.800 (0.07) | 0.656–0.944 | 0.317 |
| i-ADL-PDI | 0.571 (0.09) | 0.394–0.749 | <0.001 |

| MCI VERSUS AD | | | |
|-----------------|--------------|-------------|----------------------|
| | AUC (S.E.) | 95% CI | P-VALUE ^a |
| NAT total score | 0.809 (0.06) | 0.674–0.944 | |
| NAT task 1 | 0.753 (0.07) | 0.600–0.905 | |
| NAT task 2 | 0.613 (0.09) | 0.436–0.789 | |
| NAT task 3 | 0.813 (0.07) | 0.673–0.952 | |
| a-ADL-DI | 0.739 (0.07) | 0.584–0.893 | 0.424 |
| a-ADL-CDI | 0.814 (0.06) | 0.679–0.949 | 0.062 |
| a-ADL-PDI | 0.539 (0.09) | 0.357–0.721 | 0.022 |
| i-ADL-DI | 0.705 (0.08) | 0.538–0.872 | 0.291 |
| i-ADL-CDI | 0.808 (0.06) | 0.672–0.943 | 0.989 |
| i-ADL-PDI | 0.608 (0.09) | 0.431–0.784 | 0.073 |

| HC VERSUS AD | | | |
|-----------------|--------------|-------------|----------------------|
| | AUC (S.E.) | 95% CI | P-VALUE ^a |
| NAT total score | 1.000 (0.00) | 1.000–1.000 | |
| NAT task 1 | 0.952 (0.03) | 0.880–1.000 | |
| NAT task 2 | 0.870 (0.05) | 0.763–0.978 | |
| NAT task 3 | 0.955 (0.03) | 0.886–1.000 | |
| a-ADL-DI | 0.948 (0.04) | 0.867–1.000 | 0.210 |
| a-ADL-CDI | 0.964 (0.02) | 0.913–1.000 | 0.163 |
| a-ADL-PDI | 0.674 (0.08) | 0.505–0.842 | <0.001 |
| i-ADL-DI | 0.915 (0.04) | 0.828–1.000 | 0.058 |
| i-ADL-CDI | 0.975 (0.02) | 0.918–1.000 | 0.317 |
| i-ADL-PDI | 0.674 (0.08) | 0.507–0.841 | <0.001 |



NAT: Naturalistic Action Test; a-ADL-DI: advanced activities of daily living-disability index; a-ADL-CDI: advanced activities of daily living-cognitive disability index; a-ADL-PDI: advanced activities of daily living-physical disability index; i-ADL-DI: instrumental activities of daily living-disability index; i-ADL-CDI: instrumental activities of daily living-cognitive disability index; i-ADL-PDI: instrumental activities of daily living-physical disability index.

^ap-values indicate the significant difference between the indices and the total score of the NAT. Differences were calculated by comparing ROC curves with the method of DeLong *et al.* (1988).

First, the convergent validity was high, with strong significant correlations between the total score of the NAT and the a-ADL-DI, a-ADL-CDI, and i-ADL-CDI, and a moderate significant correlation with the i-ADL-DI. Second, the concurrent validity showed that both the NAT and the a- and i-ADL tools were able to provide an accurate diagnostic differentiation between HCs, MCI, and AD. Neither the AUC of the NAT total score nor the AUC of the a- and i-ADL tools showed significantly better accuracy. Consequently,

it can be assumed that the a- and i-ADL tools and the NAT are equally able to assess everyday functioning, and that these tools have analogous accuracy in differentiating between HCs, MCI, and AD.

Until now, performance-based evaluations have been considered more likely to detect deficits in everyday functioning in persons with cognitive disorders than report-based scales (Puente *et al.*, 2014). However, this study indicates that a report-based measure might be equal

to a performance-based measure. This contradicts prior studies showing different estimates, or at most medium correlations between report-based and performance-based measures (Schmitter-Edgecombe *et al.*, 2014; Rycroft *et al.*, 2017). Possibly, the strong correlation between the a- and i-ADL tools and the NAT might be explained by important differences between the a- and i-ADL tools and other report-based methods. By conducting a semi-structured interview, the a- and i-ADL tools use a different assessment approach to many other report-based methods for which the scoring is solely based on the success or failure of completing a task or on the presence or absence of a skill (Rodakowski *et al.*, 2014). Moreover, the a- and i-ADL tools offer the possibility of taking causes of limitations into account, which is crucial in determining the extent to which cognitive decline is responsible for functional impairment (Rockwood, 2007; Wadley *et al.*, 2008; De Vriendt *et al.*, 2015). Through discriminating and evaluating reasons of limitations, the a- and i-ADL tools can detect similar limitations in performance as when observing everyday functioning using a performance-based tool. Other report-based instruments, such as the Disability Assessment in Dementia (DAD) – 6 (de Rotrou *et al.*, 2012), the Functional Activities Questionnaire (FAQ) (Pfeffer *et al.*, 1982), and the 18-item AD Cooperative Study/ADLs Inventory for patients with MCI (ACDC-MCI-ADL-18) (Pedrosa *et al.*, 2010), do not take reasons of impairment into account, or focus only on executive components of activities. Also the NAT does not record other than cognitive reasons of impairment.

The a- and i-ADL tools and the NAT take different approaches in evaluating ADL, which consequently results in gathering other information on everyday functioning. The NAT records errors in performance and takes those errors into account in the total score. This provides unique and useful information that cannot be derived by interviewing an individual with the a- and i-ADL tools. On the other hand, the a- and i-ADL tools might have greater content validity by only taking those activities into account that are relevant for an individual, in contrast to the NAT. Wrapping a present and packing a lunch box and a schoolbag are activities not embedded in the usual daily routine of older persons and are especially difficult for persons with cognitive disorders (Gold, 2012). Even individuals with MCI lose accuracy and efficiency in completing activities outside their typical routine (De Vriendt *et al.*, 2013; Seelye *et al.*, 2013). This might explain why, in our study, task 2 of the NAT could not discriminate between

MCI and AD since both groups performed equally worse on this task. In addition, knowledge of being monitored for errors in performance also increase cognitive demands on participants (Seelye *et al.*, 2013), which might result in worse performance. Therefore, only evaluating relevant activities and listening explicitly to the subjective experiences of an individual pursues a client-centered approach not only of benefit diagnostically but also facilitates more appropriate therapy, better health outcomes, and greater patient satisfaction (Stewart *et al.*, 2011).

Since this study found a similar diagnostic accuracy for the NAT and the a- and i-ADL tools, one could argue that it does not make any difference which instrument is used in clinical practice. However, choosing one or another method depends on the clinical setting and the purpose of the diagnostic procedure. As time and resources are often scarce in health services, it might be recommended to start with the feasible a- and i-ADL tools since they take the least time to administer, offer greater diagnostic accuracy than other report-based measures such as the Lawton scale (Cornelis *et al.*, 2017). Subsequently, a performance-based method such as the NAT can be used. As suggested by Bruce *et al.* (2016), the NAT may be of particular interest to increase confidence in a clinical diagnosis or in situations in which there is no informant history available or there is doubt regarding the accuracy of the history. In this way, the NAT may be used in addition to the a- and i-ADL tools, and adds unique diagnostic information that might be missing when only the report-based approach is applied. By using multiple methods, the sensitivity of a diagnosis will certainly be maximized (Hunsley and Meyer, 2003).

This study has the strength of using different methods of assessment in the same sample, which mitigates the potential risk of comparing measures across different samples. Only few studies have done this. Nevertheless, it should be noted that this study also has some weaknesses. First of all, the sample is relatively small, which results in low statistical power, although the sample size is comparable to similar recent studies studying the psychometric value of the NAT (Jung *et al.*, 2013; Bruce *et al.*, 2016). Second, a measurement bias might have occurred in reporting ADLs. For HCs, only self-report was used since prior research suggested that self-report evaluations are generally accurate indicators of ADLs for cognitively healthy older persons, demonstrating insight into their functional abilities (Farias *et al.*, 2005; Suchy *et al.*, 2011). For patients with MCI and AD, on the other hand, a reliable informant was questioned since this is generally preferred in order to control over- or

under-estimation of functional abilities. Although each informant provided accurate information about the daily life of the patients with MCI and AD, we could not objectively determine how well patients with MCI and AD were known to the informants. Furthermore, we could not rule out that the informants were not mildly cognitively impaired themselves. However a previous study (De Vriendt *et al.*, 2013) indicated a strong patient-proxy agreement of the a-ADL tool, this might still have influenced the results. Nevertheless, this resembles clinical reality in which healthcare professionals also have to work with the information that is available. Third, similar to the study of Bruce *et al.* (2016), only the overall scores of the NAT were used. We did not investigate the type of errors while performing the NAT as we especially wanted to compare the AUC of the total score with the indices of the a- and i-ADL tools. However, it might be interesting to correlate the number and type of errors in the NAT with the indices of the a- and i-ADL tools in the groups of HCs, MCI, and AD in order to investigate further an effective evaluation of functional decline in ADLs.

In conclusion, this study showed that the ability of the report-based a- and i-ADL tools and the performance-based NAT to make a diagnostic differentiation between HCs, MCI, and AD is not significantly different. Both methods have strong concurrent and convergent validity, and appear to be valid and reliable assessments of ADLs, with similar discriminatory power in diagnosing cognitive disorders in older persons.

Ethical statement

The Ethical Committee of the Universitair Ziekenhuis Brussel approved this study (B.U.N. 143201421290). All data were collected in accordance with the ICH-GCP guidance and the Helsinki Declaration. All participants and informants gave written informed consent.

Conflict of interest

None.

Description of authors' roles

E. Cornelis designed the study, collected the data, carried out the statistical analysis, and wrote the paper. E. Gorus designed the study, was responsible for the statistical design of the study, supervised the statistical analysis, and assisted writing the paper. K. Van Weverbergh collected the data and assisted

in analyzing the data. I. Beyer supervised collecting the data, designing the study, and analyzing the data. P. De Vriendt designed the study, was responsible for the statistical design of the study, supervised the statistical analysis, and assisted writing the paper.

Supplementary material

To view supplementary material for this article, please visit <https://doi.org/10.1017/S1041610218000327>

References

- Allen, C. K., Austin, S. L., David, S. K., Earhart, C. A., McCraith, D. B. and Riska-Williams, L. (2007). *Allen Cognitive Level Screen-5 (ACLS-5) and Large Allen Cognitive Level Screen-5 (LACLS-5)*. Camarillo, CA, USA: ACLS and LACLS Committee.
- Andresen, E. M. (2000). Criteria for assessing the tools of disability outcome research. *Archives of Psychiatric Medicine and Rehabilitation*, 81, S15–20.
- Anstey, K. J. *et al.* (2013). Characterizing mild cognitive disorders in the young-old over 8 years: prevalence, estimated incidence, stability of diagnosis, and impact on iADLs. *Alzheimer's & Dementia*, 9, 640–648.
- Bean, J. F., Olveczky, D. D., Kiely, D. K., LaRose, S. I. and Jette, A. M. (2011). Performance-based versus patient-reported physical function: what are the underlying predictors? *Physical Therapy*, 91, 1804–1811.
- Bruce, I., Ntholang, O., Crosby, L., Cunningham, C. and Lawlor, B. (2016). The clinical utility of naturalistic action test in differentiating mild cognitive impairment from early dementia in memory clinic. *International Journal of Geriatric Psychiatry*, 31, 309–315.
- Cornelis, E., Gorus, E., Beyer, I., Bautmans, I. and De Vriendt, P. (2017). Early diagnosis of mild cognitive impairment and mild dementia through basic and instrumental activities of daily living: development of a new evaluation tool. *PLoS Medicine*, 14, Article no. e1002250.
- de Rotrou, J. *et al.* (2012). DAD-6: a 6-Item version of the disability assessment for dementia scale which may differentiate Alzheimer's disease and mild cognitive impairment from controls. *Dementia and Geriatric Cognitive Disorders*, 33, 210–218.
- De Vriendt, P., Mets, T., Petrovic, M. and Gorus, E. (2015). Discriminative power of the advanced activities of daily living (a-ADL) tool in diagnosis of mild cognitive impairment in an older population. *International Psychogeriatrics*, 22, 1–19.
- De Vriendt, P., Gorus, E., Cornelis, E., Bautmans, I., Petrovic, M. and Mets, T. (2013). The advanced activities of daily living: a tool allowing the evaluation of subtle functional decline in mild cognitive impairment. *The Journal of Nutrition, Health and Aging*, 17, 64–71.
- DeLong, E. R., DeLong, D. and Clarke-Pearson, D. L. (1988). Comparing the areas under two or more correlated

- receiver operating characteristics curves: a nonparametric approach. *Biometrics*, 44, 837–845.
- Dubois, B., Padovani, A., Scheltens, P., Rossi, A. and Dell’Agnello, G.** (2016). Timely diagnosis for Alzheimer’s disease: a literature review on benefits and challenges. *Journal of Alzheimer’s Disease*, 49, 617–631.
- Farias, S. T., Mungas, D. and Jagust, W.** (2005). Degree of discrepancy between self and other-reported memory and everyday functioning by cognitive status: dementia, mild cognitive impairment and healthy elders. *International Journal of Geriatric Psychiatry*, 20, 827–834.
- Fauth, E. B., Schwartz, S., Tschanz, J., Ostbye, T., Corcoran, C. and Norton, M. C.** (2013). Baseline disability in activities of daily living predicts dementia risk even after controlling for baseline global cognitive ability and depressive symptoms. *International Journal of Geriatric Psychiatry*, 28, 597–606.
- Fieo, R., Austin, E. J., Starr, J. M. and Deary, I. J.** (2011). Calibrating ADL-IADL scales to improve measurement accuracy and to extend the disability construct into the preclinical range: a systematic review. *BMC Geriatrics*, 11, 42.
- Fisher, A.** (1999). *Assessment of Motor and Process Skills*. Fort Collins, CO: Three Star Press.
- Folstein, M. F., Folstein, S. E. and McHugh, P. R.** (1975). “Mini-mental state”. A practical method for grading the cognitive state of patients for the clinician. *Journal of Psychiatric Research*, 12, 189–198.
- Gallagher, D. et al.** (2010). Detecting prodromal Alzheimer’s disease in mild neuropsychological, cognitive impairment: utility of the CAMCOG and other predictors. *International Journal of Geriatric Psychiatry*, 25, 1280–1287.
- Giovannetti, T., Libon, D., Buxbaum, L. J. and Schwartz, M. F.** (2002a). Naturalistic action impairments in dementia. *Neuropsychologia*, 40, 1220–1232.
- Gold, D. A.** (2012). An examination of instrumental activities of daily living assessment in older adults and mild cognitive impairment. *Journal of Clinical and Experimental Neuropsychology*, 34, 11–34.
- Hugo, J. and Ganguli, M.** (2014). Dementia and cognitive impairment: epidemiology, diagnosis and treatment. *Clinics in Geriatric Medicine*, 30, 421–442.
- Hunsley, J. and Meyer, G.** (2003). The incremental validity of psychological testing and assessment: conceptual, methodological, and statistical issues. *Psychological Assessment*, 15, 446–455.
- Jekel, K. et al.** (2015). Mild cognitive impairment and deficits in instrumental activities of daily living: a systematic review. *Alzheimer’s Research & Therapy*, 7, 17.
- Jung, M. A., Kim, H., Kang, Y. J. and Kim, Y. J.** (2013). Development of a modified naturalistic action test for Korean patients with impaired cognition. *Annals of Rehabilitation Medicine*, 37, 57–65.
- Katz, S.** (1963). Studies of illness in the aged. The index of ADL: a standardized measure of biological and psychosocial function. *JAMA*, 185, 914–919.
- Lawton, M. P. and Brody, E. M.** (1969). Assessment of older people: self-maintaining and instrumental activities of daily living. *Gerontologist*, 9, 179–186.
- Marshall, G. A., Amariglio, R., Sperling, R. A. and Rentz, D. M.** (2012). Activities of daily living: where do they fit in the diagnosis of Alzheimer’s disease? *Neurodegenerative Disease Management*, 2, 483–491.
- McKhann, G. M.** (1984). Clinical diagnosis of Alzheimer’s disease: report of the NINCDS-ADRDA working group under the auspices of department of health and human services task force on Alzheimer’s disease. *Neurology*, 34, 939–944.
- McKhann, G. M. et al.** (2011). The diagnosis of dementia due to Alzheimer’s disease: recommendations from the national institute on aging–Alzheimer’s association workgroups on diagnostic guidelines for Alzheimer’s disease. *Alzheimer’s & Dementia*, 7, 263–269.
- Nielsen, L. M., Kirkegaard, H., Ostergaard, L. H., Bovbjerg, K., Breinholt, K. and Maribo, T.** (2016). Comparison of self-reported and performance-based measures of functional ability in elderly patients in an emergency department: implications for selection of clinical outcome measures. *BMC Geriatrics*, 16, 199.
- Pedrosa, H. et al.** (2010). Functional evaluation distinguishes MCI patients from healthy elderly people – the ADCS/MCI/ADL scale. *Journal of Nutrition Health and Aging*, 14, 703–709.
- Petersen, R. C.** (2004). Mild cognitive impairment as a diagnostic entity. *Journal of Internal Medicine*, 256, 240–246.
- Petersen, R. C., Caracciolo, B., Brayne, C., Gauthier, S., Jelic, V. and Fratiglioni, L.** (2014). Mild cognitive impairment: a concept in evolution. *Journal of Internal Medicine*, 275, 214–228.
- Pfeffer, R. I., Kurosaki, T. T., Harrah, C. H., Chance, J. M. and Filos, S.** (1982). Measurement of functional activities in older adults in the community. *Journal of Gerontology*, 37, 323–329.
- Puente, A. N., Terry, D. P., Faraco, C. C., Brown, L. C. and Miller, L. S.** (2014). Functional impairment in mild cognitive impairment evidenced using performance-based measurement. *Journal of Geriatric Psychiatry and Neurology*, 27, 253–258.
- Reppermund, S. et al.** (2016). Performance-based assessment of instrumental activities of daily living: validation of the Sydney Test of Activities of Daily Living in Memory Disorders (STAM). *Journal of the American Medical Directors Association*, 18, 117–122.
- Reuben, D. B., Laliberte, L., Hiris, J. and Mor, V.** (1990). A hierarchical exercise scale to measure function at the Advanced Activities of Daily Living (AADL) level. *Journal of the American Geriatrics Society*, 38, 855–861.
- Rockwood, K.** (2007). The measuring, meaning and importance of activities of daily living (ADLs) as an outcome. *International Psychogeriatrics*, 19, 467–482.
- Rodakowski, J. et al.** (2014). Can performance on daily activities discriminate between older adults with normal cognitive function and those with mild cognitive impairment? *Journal of the American Geriatrics Society*, 62, 1347–1352.
- Roedersheimer, K. M., Pereira, G. F., Jones, C. W., Braz, V. A., Mangipudi, S. A. and Platts-Mills, T. F.** (2016). Self-reported versus performance-based assessments of a simple mobility task among older adults in the emergency department. *Annals of Emergency Medicine*, 67, 151–6.

- Rycroft, S. S., Giovanetti, T., Divers, R. and Hulswit, J.** (2017). Sensitive performance-based assessment of everyday action in older and younger adults. *Neuropsychology, Development, and Cognition Section B: Aging Neuropsychology and Cognition*, 7, 1–18.
- Sager, M. A., Dunham, N. C., Schwantes, A., Mecum, L., Halverson, K. and Harlowe, D.** (1992). Measurement of activities of daily living in hospitalized elderly: a comparison of self-report and performance-based methods. *Journal of the American Geriatric Society*, 40, 457–462.
- Schmitter-Edgecombe, M., Parsey, C. and Cook, D. J.** (2014). Cognitive correlates of functional abilities in individuals with mild cognitive impairment: comparison of questionnaire, direct observation and performance-based measures. *The Clinical Neuropsychologist*, 28, 726–746.
- Schwartz, M. F., Segal, M., Veramonti, T., Ferraro, M. and Buxbaum, L. J.** (2002). The naturalistic action test: a standardised assessment for everyday action impairment. *Neuropsychological Rehabilitation*, 12, 311–339.
- Seelye, A. M., Schmitter-Edgecombe, M., Cook, D. J. and Crandall, A.** (2013). Naturalistic assessment of everyday activities and prompting technologies in mild cognitive impairment. *Journal of the International Neuropsychological Society*, 19, 442–452.
- Sestito, N., Schmidt, K., Gallo, J. L., Giovannetti, T. and Libon, D. J.** (2005). Using the Naturalistic Action Test (NAT) to assess everyday action in healthy older adults and patients with dementia. *Journal of the International Neuropsychological Society*, 11, 90–91.
- Sikkes, S. A. and Rotrou, J. D.** (2014). A qualitative review of instrumental activities of daily living in dementia: what's cooking? *Neurodegenerative Disease Management*, 4, 393–400.
- Stewart, M. et al.** (2011). Instrumental activities of daily living among community-dwelling older adults: discrepancies between self-report and performance are mediated by cognitive reserve. *Journal of Clinical and Experimental Neuropsychology*, 33, 92–100.
- Suchy, Y., Kraybill, M. L. and Franchow, E.** (2011). Instrumental activities of daily living among community-dwelling older adults: discrepancies between self-report and performance are mediated by cognitive reserve. *Journal of Clinical and Experimental Neuropsychology*, 33, 92–100.
- Wadley, V. G., Okonkwo, O., Crowe, M., and Ross-Meadows, L. A.** (2008). Mild cognitive impairment and everyday function: evidence of reduced speed in performing instrumental activities of daily living. *American Journal of Geriatric Psychiatry*, 16, 416–424.
- Wesson, J., Cemson, L., Brodaty, H. and Reppermund, S.** (2016). Estimating functional cognition in older adults using observational assessments of task performance in complex everyday activities: a systematic review and evaluation of measurement properties. *Neuroscience and Biobehavioral Reviews*, 68, 335–360.
- World Health Organization.** (2001). *International Classification of Functioning, Disability and Health (ICF)*. Geneva, Switzerland: World Health Organization.

The unmet support needs of carers of older Australians: prevalence and mental health

Jeromey B. Temple¹ and Briony Dow^{1,2}

¹Demography and Ageing Unit, Melbourne School of Population and Global Health, University of Melbourne, Melbourne, Victoria, Australia

²National Ageing Research Institute (NARI), Melbourne, Victoria, Australia

ABSTRACT

Background: Population aging places greater demands on the supply of informal carers. The aims of this study were to examine (1) the types of unmet support needs of carers of older Australians and (2) the association of unmet needs with mental health.

Methods: Utilizing new data from the 2015 Australian Bureau of Statistics (ABS) Survey of Disability, Ageing and Carers, we calculated the prevalence of carers experiencing specific and multiple unmet needs for support, using single and multiple item measures. Logistic regression models were fitted to examine the association between unmet needs and psychological distress (using the Kessler psychological distress scale), once demographic and health factors were controlled for.

Results: In 2015, 35% of carers of older Australians cited at least one unmet need for support. Among this group, almost two-thirds cited multiple unmet support needs (64.7%). The most prevalent types of unmet needs included financial (18%), physical (13%), and emotional support (12%), as well as additional respite care and support to improve carer health (12%). After controlling for demographic and health characteristics of the carer, having any unmet need for support increased the odds of psychological distress by twofold (OR = 2.20, 95% CI = 1.65, 2.94). With each successive unmet need for support, the odds of psychological distress increased 1.37 times (OR = 1.36, 95% CI = 1.22, 1.54). Those who had received assistance with care, but required further support were 1.95 times more likely (OR = 1.95, 95% CI = 1.17, 3.24) to be in distress and those who had not received care assistance were about 2.4 times more likely (OR = 2.38 95% OR = 1.56, 3.62) to be in distress relative to those with no unmet need.

Conclusions: Addressing unmet support needs of carers is important, not only for the planning of services for carers in an aging population, but also because of the association between unmet support needs and carers mental health.

Key words: caring, informal care, aging, psychological distress

Introduction

With the speed of population aging hastening across most high-income countries, the care needs of the growing number of older citizens has been an important priority for policy-makers. Consistent with the preferences of older persons, successive Australian governments have sought to assist older people to receive care in their own homes, rather than institutions since the 1980s (Dow *et al.*, 2013). One of the factors key to the success of

this strategy is the large pool of informal carers in Australia, providing unpaid caring activities, estimated to account for 80% of the care of older people in Australia (Productivity Commission, 2011). In 2015, 2.7 million people provided informal care with an estimated economic value of \$60 billion (Deloitte Access Economics, 2015; Australian Bureau of Statistics [ABS], 2016b). The United Kingdom and USA also rely significantly on informal carers to care for older persons. Six and a half million people in the United Kingdom are carers, with 44% caring for a parent or grandparent (Carers UK, 2015) and 34.2 million Americans provide unpaid care to a person aged over 50 years (National Alliance for Caregiving, 2015).

Although informal caring is mostly unpaid, a number of government programs and payments are in place to assist Australian carers. Carers

Correspondence should be addressed to: Jeromey B. Temple, Associate Professor, Demography and Ageing Unit, Melbourne School of Population and Global Health, University of Melbourne, 207 Bouverie St, Melbourne, VIC 3010 Australia. Phone: +61-3-90359900. Email: Jeromey.Temple@unimelb.edu.au. Received 21 Aug 2017; revision requested 24 Oct 2017; revised version received 20 Feb 2018; accepted 26 Feb 2018. First published online 25 May 2018.

can access respite care and several types of carer payment, including an income support payment for those who are unable to work in paid employment due to their caring role through the government's Carer Gateway (Australian Government, 2017). There are carer advocacy and support services in every state and territory through which carers can gain access to services such as counselling and support groups (Carers Australia, 2017). Carers in Australia also have the right to request flexible working arrangements enshrined in legislation and this request cannot be reasonably refused by employers (Victorian Equal Opportunity and Human Rights Commission, 2012). There are similar arrangements in place for carers in the United Kingdom, but in the USA there is no income support payment for carers (Agingcare, 2017).

Despite these government programs, research has shown that many carers have unmet needs for further support. Some carers require support and education on practical skills to assist with their caregiving role and access to support services is oftentimes limited due to complex eligibility criteria (Essue *et al.*, 2010). This is complicated further by the low levels of assistance to primary caregivers and awareness of many existing community services for carers and their recipients (Lucke *et al.*, 2008; Vecchio, 2008b). Users of different support services may also have differing levels of unmet support needs. In an examination of respite service use in 2003, respite care users had considerable unmet demands for financial assistance, whereas non-respite care users had unmet needs to improve their own well-being (Vecchio, 2008a). More generally, stressors associated with caregiving may impact upon the carer, requiring additional needs, including health and emotional support (O'Connell *et al.*, 2003; Horner and Boldy, 2008). This issue of unmet emotional support is important, as there is considerable evidence that carers are at increased risk of adverse mental health, including depression (Gaugler *et al.*, 2004; Cummins *et al.*, 2007; Dahlberg *et al.*, 2007; Neri *et al.*, 2012).

Despite the considerable contribution of the above studies to our understanding of carer needs, there remains a gap in studies of the types of needs of carers, specifically among those who care for older Australians and those who have multiple unmet support needs. With the availability of new nationally representative data, we seek to answer two questions: (1) What is the prevalence and type of unmet support needs of those who care for older Australians? (2) Is there an association between the carers' unmet support needs and mental health?

Methods

Data

Data for this study are from the 2015 Survey of Disability, Ageing and Carers (SDAC) conducted between July and December 2015. Three populations were sampled using multi-stage sampling techniques. These consisted of persons living in private dwellings, in self-care retirement villages, and in cared accommodation. The module on carers' unmet needs was administered to primary carers living in households. Primary carers were identified through a series of screening questions. As defined by the ABS, primary carers "provide(s) the most informal assistance, in terms of help or supervision, to a person with one or more disabilities, with one or more of the core activities of mobility, self-care or communication" (ABS, 2016a). The ABS has the authority to collect these data under the Census and Statistics Act 1905, and has collected eight cross sections since 1981. Respondents were neither paid for participation nor were any other inducements offered.

Of 31,957 households originally contacted, 25,555 fully responded, yielding a response rate of 80%. Of these persons living in households, 2,421 were primary carers who filled out the unmet needs questionnaire. Of this group, we omit 340 participants where the primary carer did not answer questions pertaining to unmet needs. This left a final sample size of 2,081 primary carers. Within this group approximately 18% care for children ($n = 382$), 20% care for person in the primary working ages ($n = 420$), and 61% care for older Australians aged 55 years and over ($n = 1,274$). This later group of carers are the focus of this study, representing over 60% of primary carers. We used the cut-off age of 55 years because from age 55, persons born before 1960 can access private superannuation and retire in Australia. The average age of primary carers (of recipients of any age) in Australia is 55 years, with two-thirds being female and the majority caring for a family member (ABS, 2016b).

Measures

In the self-completed carer questionnaire, primary carers were asked "Do you feel you need more support or an improvement in your situation to aid in your role as a carer?" A follow up question was then asked "Which of the following would assist you in your role as a carer?" A list of possible responses was then provided, including the following:

1. more respite care,
2. more financial assistance,
3. more physical assistance,

4. more emotional support,
5. an improvement in my own health,
6. more aids/equipment to help me assist in my role as a carer,
7. more courses available on how to care for persons with particular disabilities,
8. more training in correct use of equipment,
9. more training in correct methods of lifting to prevent injury to myself, and
10. none of the above.

For those who answered multiple needs, they were asked to nominate which would “most assist you in your role as carer?” Herein, this later measure is referred to as the single item measure.

For the first time in 2015, the SDAC collected measures of psychological distress. The specific measure of psychological distress used was Kessler’s K10. The K10, although not a diagnostic tool, is a widely used screening instrument for psychological distress based on the respondent’s emotional state in the four weeks prior to the interview (Anderson *et al.*, 2013; ABS, 2016a). This enables an analysis of whether psychological distress is associated with unmet carer support needs. Following other examples in the literature, we indicate a “high” or “very high” score on Kessler’s measure as indicating psychological distress (Anderson *et al.*, 2013).

Statistical model and estimation of variance

To examine differences in the prevalence of unmet carer needs and psychological distress we present weighted tests of proportions, with a 95% critical value. To examine the association between unmet carer needs and psychological distress we fitted logistic regression models. Using the raw logit coefficients, we calculated odds ratios (OR), which measure the change in the odds of experiencing psychological distress given a change in unmet carer needs, once all other factors in the model are controlled for.

Variables were entered into the regression and improvement to model fit assessed using the Bayesian Information Criteria following the Raftery (1995) procedure. With all models specified, we checked the conditioning of the matrix of independent variables to investigate any collinearity influence (Belsley *et al.*, 1980). The condition numbers and variance inflation factors were small to provide support for the model specification. Final goodness-of-fit for the logistic regression models was confirmed using the Hosmer and Lemeshow test (Hosmer *et al.*, 1997).

Due to the complex survey design, adjustments are necessary to generate correct variance estimates. The SDAC includes 60 replicate weights

on the data file to adjust for sample design and non-response. Utilizing an algorithm developed by Winter (2008), we employed the unstratified delete-one jackknife method to make the necessary replicate adjustments (Wolfer, 1985). All analyses were conducted using Stata 15 (Stata Corporation, 2017).

Results

Table 1 displays the differences in prevalence of carer unmet support needs using the single item (most assist you) and multiple item measure (any need for assistance). Regardless of the measure used, about one-third of the primary carers of older persons have unmet demands for support (34.6%). There is considerable variation in the prevalence of additional support requested by carers according to the measure used (Table 1). Relying only on the single item measure significantly underrepresents the full demand for a number of sources of support. For example, using the single item measure, about 9% of carers report “more financial assistance” compared with almost one in five carers responding to the multiple item measure (17.7%). Similarly, only 3% report a need for emotional support on the single item measure, but about 12% of carers indicate this unmet need on the multiple item measure (11.9%).

Focusing on the multiple item measure, financial assistance is the most heavily cited among carers as an unmet need (17.7%). Half of all carers with unmet needs cited this unmet support need (49.7%). In addition, over 10% of carers report additional physical assistance (12.7%), support with improving their own health (12.1%), more emotional support (11.9%), and respite care (11.6%) as key support needs to improve their caring role. Around 6% of carers report a need for additional aids or equipment (6.3%) or courses on caring for persons with particular disabilities (5.8%).

Using the multiple item measure, we generate a variable measuring the number of types of unmet needs of carers. In Table 2, we tabulate the types of unmet needs by number of types of unmet needs. This table includes the population of those who report any unmet need ($n = 445$). Of those with unmet needs, 35% of carers have one unmet need only, 43% have two or three unmet needs and about one in five have four or more unmet needs (21.5%). In total, almost two-thirds of carers with unmet needs have multiple unmet needs (64.7%), representing 22% of all primary carers of older Australians.

As expected, the proportions reporting across all unmet support need types increase with total

Table 1. Prevalence of carer unmet support needs, by type, single and multiple measures, 2015

| | SINGLE | | MULTIPLE | |
|--|---------|------------|----------|------------|
| | MEASURE | 95% C.I. | MEASURE | 95% C.I. |
| No additional support required | 65.4 | 62.4, 67.6 | 65.4 | 62.4, 67.6 |
| Additional support required | 34.6 | 32.4, 37.6 | 34.6 | 32.4, 37.6 |
| Source of support required | | | | |
| More respite care | 5.8 | 4.5, 7.1 | 11.6 | 10.1, 13.7 |
| More financial assistance | 9.3 | 7.8, 10.9 | 17.7 | 15.4, 19.5 |
| More physical assistance | 4.1 | 3.3, 5.6 | 12.7 | 11.1, 14.8 |
| More emotional support | 3.1 | 2.5, 4.5 | 11.9 | 10.5, 14.1 |
| An improvement in your own health | 3.4 | 2.4, 4.4 | 12.1 | 10.9, 14.5 |
| More aids/equipment to help you assist in your role as a carer | 1.7 | 0.9, 2.2 | 6.3 | 5.2, 7.9 |
| More courses available on how to care for persons with particular disabilities | 1.3 | 1.0, 2.4 | 5.8 | 5.0, 7.7 |
| More training on correct use of equipment | 0.1 | 0.0, 0.6 | 1.94 | 1.4, 3.0 |
| More training in correct methods of lifting to prevent injury to carer | 0.3 | 0.1, 0.8 | 3.3 | 2.7, 4.7 |
| None of the above | 1.7 | 1.3, 2.8 | 1.72 | 1.3, 2.8 |
| Source of support not answered | 3.7 | 2.8, 4.9 | 0.2 | 0.0, 0.7 |
| Total | 34.6 | | | |

Notes: 95% CI: 95% confidence interval for logit proportions. All percentages are weighted to account for survey design. Unweighted confidence intervals reported for transparency.

Table 2. Prevalence of carer unmet support needs, by type and number of unmet needs, 2015

| | NUMBER OF UNMET NEEDS | | | |
|--|-----------------------|---------|---------|-------|
| | 1 ^a | 2–3 | 4+ | All |
| More respite care | 15.1 | 32.2*** | 66.8*** | 33.7 |
| More financial assistance | 33.4 | 51.2*** | 79.8*** | 49.7 |
| More physical assistance | 7.1 | 47.7*** | 63.9*** | 36.6 |
| More emotional support | 9.7 | 35.3*** | 72.9*** | 34.8 |
| An improvement in your own health | 10.9 | 39.6*** | 65.2*** | 36.0 |
| More aids/equipment to help you assist in your role as a carer | 4.6 | 12.4*** | 52.1*** | 18.4 |
| More courses available on how to care for persons with particular disabilities | 3.1 | 14.4*** | 47.9*** | 17.8 |
| More training on correct use of equipment | 0.0 | 1.7* | 22.7*** | 5.8 |
| More training in correct methods of lifting to prevent injury to carer | 0.7 | 4.2** | 34.4*** | 10.1 |
| None of the above | 14.1 | 0.0 | 0.0 | 5.4 |
| Source of support not answered | 1.6 | 0.0 | 0.0 | 0.7 |
| <i>n</i> | 159 | 188 | 98 | 445 |
| % of carers with unmet needs | 35.3 | 43.2 | 21.5 | 100.0 |
| % of all carers | 12.2 | 15.0 | 7.4 | 34.6 |

Notes: All: All persons with unmet support needs; percentages weighted. Tests of percentages unweighted.

^aComparison category for test of percentages is 1 unmet need.

****p* < 0.001.

***p* < 0.01.

**p* < 0.05.

unmet needs. For example, 7% of those with one unmet need require physical assistance, compared with half of those with two or three unmet needs (47.7%) and almost two-thirds of those with four or more unmet needs (63.9%). Of those with multiple needs, considerable proportions of these groups cite financial, physical, and emotional support.

However, those with four or more needs also have considerably high demand for training and courses compared with those who have fewer unmet needs. Half of those with four or more unmet needs have unmet demand for “more aids/equipment” (52.1%) or “courses available on how to care for persons with particular disabilities” compared with

Table 3. Characteristics of primary carers of older Australians and unmet support needs, 2015

| | | ANY UNMET NEEDS | | TOTAL N | TOTAL CARERS (%) |
|---|--------------------|-----------------|---------|---------|------------------|
| | | YES (%) | NO (%) | | |
| Age | <44 | 11.1 | 16.3*** | 134 | 12.9 |
| | 45–74 | 70.6 | 73.4† | 915 | 71.6 |
| | 75+ | 18.2 | 10.3*** | 206 | 15.5 |
| Gender | Male | 37.3 | 33.0*** | 440 | 35.8 |
| | Female | 62.7 | 67.0† | 815 | 64.2 |
| Country of birth | Australia | 73.0 | 70.2*** | 904 | 72.0 |
| | MESB | 10.5 | 11.6 | 148 | 10.9 |
| | Other | 16.5 | 18.2 | 203 | 17.1 |
| Social marital status | Married | 74.0 | 61.4*** | 881 | 69.7 |
| | Sep/Wid/Div | 12.7 | 19.4** | 194 | 15.1 |
| | Never married | 13.2 | 19.1* | 180 | 15.3 |
| Region of residence | Major city | 66.0 | 69.1*** | 802 | 67.1 |
| | Inner regional | 21.7 | 21.9 | 270 | 21.8 |
| | Other | 12.2 | 9.0 | 183 | 11.1 |
| Labor force status | Employed | 33.2 | 38.2*** | 424 | 35.0 |
| | Unemployed | 1.1 | 2.0 | 19 | 1.4 |
| | NILF | 65.6 | 59.8* | 812 | 63.6 |
| Education care recipient spouse | Degree or above | 17.5 | 24.8*** | 248 | 20.0 |
| | Certificate | 30.0 | 35.0* | 385 | 31.8 |
| | School only | 37.4 | 29.8** | 454 | 34.4 |
| | Less than 8th form | 13.4 | 9.5* | 146 | 12.0 |
| | Undetermined | 1.8 | 1.9 | 22 | 1.8 |
| Carer co-resident | Yes | 51.6 | 40.6 | 652 | 47.8 |
| | No | 48.4 | 29.4*** | 603 | 52.2 |
| Carers' number of long-term health conditions | Yes | 73.2 | 64.8*** | 890 | 70.3 |
| | No | 26.8 | 35.2*** | 365 | 29.7 |
| Duration of care | 0 | 26.1 | 26.3*** | 302 | 26.2 |
| | 1–2 | 23.8 | 35.5 | 453 | 35.7 |
| | 3–4 | 23.8 | 24.3 | 304 | 24.2 |
| | 4+ | 14.33 | 13.95 | 196 | 15.6 |
| | Missing | 1.5 | 1.4 | 19 | 1.4 |
| Receipt of care assistance | <1 | 5.9 | 5.6*** | 76 | 5.8 |
| | 1–9 | 63.4 | 63.8 | 788 | 63.5 |
| Receipt of care assistance | 10+ | 29.2 | 29.3 | 372 | 29.2 |
| | Yes | 59.4 | 51.4*** | 744 | 56.7 |
| Receipt of care assistance | No | 40.6 | 48.6*** | 511 | 43.4 |

Notes: MESB: main English-speaking countries; NILF: not in the labor force; %Carers: the percentage of carers in each demographic group.

***p < 0.001.

**p < 0.01.

*p < 0.05.

†p < 0.1.

under 5% of those with one unmet need and under 15% of those with two or three unmet needs.

The results above indicate a considerable proportion of carers report unmet and multiple unmet support needs, and that those with higher numbers of needs report demand for support across a range of need types. An important question, therefore, is can unmet needs place additional stressors upon carers influencing their mental

health? As these data are cross sectional, we cannot infer causation, but we can investigate whether there are differences and an association between increasing likelihood of unmet needs with increased likelihood of psychological distress.

Before turning to the multivariate analyses, results in Table 3 illustrates the differences in demographic characteristics and care details of carers tabulated by whether they have unmet

support needs. Consistent with evidence on carers of persons of any age reported by the ABS, about two-thirds of carers of older Australians are female (64%) (ABS, 2016b). About 70% of this group were born in Australia (72%), are married (69.7%), living in a major city (67.1%), co-resident with the recipient of care (70.3%), and aged between 45 and 74 years (71.6%).

Comparing across unmet support need status, those with unmet needs tend to be slightly older, married and not in the labor force and with lower levels of education compared to those with no unmet support needs. It is not surprising then that about 52% of those with unmet support needs are caring for a spouse, compared with 40% of those with no unmet needs. Those with no unmet support needs are also slightly more likely (48.6%) to have used support services compared to those with no unmet needs (40.6%). There are no meaningful differences between the groups by duration of care or the carers' number of long-term conditions.

Results in Table 4 illustrate the differences in the prevalence of psychological distress in this group of carers of older Australians (60% of all primary carers) by the demographic and care characteristics provided above. Among this group of carers, approximately one-third of those with any unmet support needs were in psychological distress (31.3%), compared with 17% of those with no unmet needs. We also observe considerable differences in the prevalence of psychological distress by the number of unmet support needs. Compared to the 17% of those with no unmet needs in distress, about 26% of those with two or more needs and over half of those with three or more needs (52.6%) report psychological distress.

To examine the association between unmet needs and psychological distress, it is important to control for demographic groups with differing prevalence rates. For example, variations in psychological distress in these data appear by age, gender, country of birth, education level, and the number of health conditions that the carer reports (Table 4).

With extensive demographic controls, the multivariate evidence strongly supports the proposition that unmet support needs of carers is associated with carers' mental health (Table 5). With control variables included, having any unmet need for support increases the odds of psychological distress twofold (Model 1: OR = 2.20, $p < 0.001$). Including a continuous measure of unmet support needs, we find that with each additional unmet support need, the odds of carer psychological distress increases 1.37 times (Model 2: OR = 1.36, $p < 0.001$). Further support is provided in Model 3, which includes a categorical variable

measuring combinations of receipt of whether the carer received any assistance to care and unmet need. Not surprisingly, there is no difference in distress between those who do not have unmet support needs classified by care assistance usage (OR = 0.98, $p < 0.10$). However, those who have received care assistance and have continuing unmet needs are about 1.95 times more likely to be in distress relative to those with no unmet needs (OR = 1.95, 95% CI = 1.17, 3.24). Those who have unmet needs and have not received care assistance are at almost 2.4 times the risk of distress (OR = 2.38, 95% CI = 1.56, 3.62).

Previous studies highlight the impact of dementia on carers' levels of stress and well-being (Gaugler *et al.*, 2004; Bertrand *et al.*, 2006; Stirling *et al.*, 2010). In the confidentialized data we utilize, the care recipient's conditions are only available for co-resident carers. That is, for primary carers living in households who have non-usual resident care recipients, the condition data is unfortunately unavailable. Restricting our sample to 890 co-resident carers, we re-estimated all models measuring a care recipient with dementia (Model 4). Unsurprisingly, carers with a care recipient who has dementia are at an almost twofold risk of distress compared to those caring for a person without dementia (OR = 1.89, 95% CI = 1.01, 3.5). Importantly, even with the additional controls for dementia and the restricted sample size, the coefficients measuring unmet needs and receipt of care assistance are highly comparable to the full sample models. That is, those who have received care assistance and have unmet needs are significantly more likely to be in distress (OR = 1.91, 95% CI = 1.02, 3.55) as are those who have unmet needs but no care service use (OR = 2.7, 95% CI = 1.70, 4.29).

Although not the purpose of the analyses, the control variables are also of substantive interest themselves. Among this group of carers of older Australians, psychological distress is highest among females (vs. males), those from a non-English speaking background (vs. Australian born), younger carers and those with low levels of education (vs. university educated). The number of health conditions of the carer is also strongly associated with the odds of psychological distress (OR = 1.39, $p < 0.001$). A range of other characteristics, including marital status, geography, income, carer relationship, and co-residence status, were tested and found not to be significant predictors of psychological distress in this sample. Polynomial terms for the number of health conditions and unmet needs, as well as various interaction terms were also not significant. Both models show absence of multicollinearity

Table 4. Characteristics of primary carers of older Australians and psychological distress, 2015

| | | DISTRESS (%) | N | CARERS (%) | |
|---|--------------------|---------------------|---------------------|------------|------|
| Measures of unmet needs | | | | | |
| Any unmet need | No | 17.3 ^a | 817 | 65.4 | |
| | Yes | 31.3 ^{***} | 438 | 34.6 | |
| Number of unmet needs | 0 | 17.3 ^a | 817 | 65.4 | |
| | 1 | 24.3 | 155 | 12.1 | |
| | 2 | 26.4 ^{**} | 186 | 15.0 | |
| | 3+ | 52.6 ^{***} | 97 | 7.5 | |
| Unmet needs × care assistance | | | | | |
| | Unmet need | Care assistance | | | |
| | No | No | 17.9 ^a | 512 | 38.8 |
| | No | Yes | 16.4 | 305 | 26.5 |
| | Yes | No | 34.6 ^{***} | 232 | 17.8 |
| | Yes | Yes | 27.9 ^{**} | 206 | 16.8 |
| Carer characteristics | | | | | |
| Age | <44 | 31.3 ^a | 134 | 12.9 | |
| | 45–74 | 22.5 | 915 | 71.6 | |
| | 75+ | 12.9 ^{***} | 206 | 15.5 | |
| Gender | Male | 17.1 ^a | 440 | 35.8 | |
| | Female | 25.0 ^{***} | 815 | 64.2 | |
| Country of birth | Australia | 21.0 ^a | 904 | 72.0 | |
| | MESB | 13.3 | 148 | 10.9 | |
| | Other | 32.8 ^{***} | 203 | 17.1 | |
| Social marital status | Married | 20.3 ^a | 881 | 69.7 | |
| | Sep/Wid/Div | 27.0 | 194 | 15.1 | |
| | Never married | 25.8 | 180 | 15.3 | |
| Region of residence | Major city | 22.6 ^a | 802 | 67.1 | |
| | Inner regional | 21.0 | 270 | 21.8 | |
| | Other | 21.8 | 183 | 11.1 | |
| Labor force status | Employed | 18.2 ^a | 424 | 35.0 | |
| | Unemployed | 36.4 | 19 | 1.4 | |
| | NILF | 24.0 [*] | 812 | 63.6 | |
| Education | Degree or above | 17.1 ^a | 248 | 20.0 | |
| | Certificate | 22.7 [*] | 385 | 31.8 | |
| | School only | 22.1 ^{**} | 454 | 34.4 | |
| | Less than 8th form | 31.5 ^{***} | 146 | 12.0 | |
| | Undetermined | 8.2 | 22 | 1.8 | |
| Care recipient spouse | Yes | 21.4 ^a | 652 | 47.8 | |
| | No | 22.9 | 603 | 52.2 | |
| Carer co-resident | Yes | 23.0 ^a | 890 | 70.3 | |
| | No | 20.2 | 365 | 29.7 | |
| Carers' number of long-term health conditions | | | | | |
| | 0 | 14.4 ^a | 302 | 26.2 | |
| | 1–2 | 15.1 | 453 | 35.7 | |
| | 3–4 | 31.3 ^{***} | 304 | 24.2 | |
| | 4+ | 38.8 ^{***} | 196 | 15.6 | |
| Duration of care (years) | | | | | |
| | <1 | 29.5 ^a | 76 | 5.8 | |
| | 1–9 | 19.6 [†] | 788 | 63.5 | |
| | 10+ | 26.5 | 372 | 29.2 | |
| | Missing | 18.7 | 19 | 1.4 | |

Notes: MESB: main English-speaking countries; NILF: not in the labor force; %Carers: the percentage of carers in each demographic group.

^aComparison category for test of proportions.

***p < 0.001.

**p < 0.01.

*p < 0.05.

†p < 0.1.

Table 5. Odds ratios from models of psychological distress, 2015

| | MODEL 1 ANY UNMET | | MODEL 2 NUMBER OF UNMET | | MODEL 3 CARE ASSIST. | | MODEL 4 DEMENTIA | |
|-------------------------------|----------------------|--------------|----------------------------|--------------|-------------------------|--------------|---------------------|--------------|
| | OR | 95% C.I. | OR | 95% C.I. | OR | 95% C.I. | OR | 95% C.I. |
| Measures of unmet needs | | | | | | | | |
| Any unmet need | | | | | | | | |
| No | a | | | | | | | |
| Yes | 2.20 | (1.65, 2.94) | | | | | | |
| Number of unmet needs | | | 1.37 | (1.22, 1.54) | | | | |
| Unmet needs × care assistance | | | | | | | | |
| Unmet need | | | | | | | | |
| No | | | | | a | | a | |
| No | | | | | 0.98 | (0.66, 1.45) | 0.89 | (0.52, 1.50) |
| Yes | | | | | 2.38 | (1.56, 3.62) | 2.7 | (1.70, 4.29) |
| Yes | | | | | 1.95 | (1.17, 3.24) | 1.91 | (1.02, 3.55) |
| Control variables | | | | | | | | |
| Age | | | | | | | | |
| <44 | a | | a | | a | | a | |
| 45–74 | 0.41 | (0.25, 0.68) | 0.43 | (0.25, 0.72) | 0.41 | (0.25, 0.68) | 0.37 | (0.18, 0.75) |
| 75+ | 0.13 | (0.07, 0.25) | 0.14 | (0.07, 0.28) | 0.13 | (0.07, 0.25) | 0.12 | (0.05, 0.27) |
| Gender | | | | | | | | |
| Male | a | | a | | a | | a | |
| Female | 1.72 | (1.14, 2.60) | 1.71 | (1.12, 2.59) | 1.73 | (1.14, 2.61) | 2.13 | (1.29, 3.53) |
| Country of birth | | | | | | | | |
| Australia | a | | a | | a | | a | |
| MESB | 0.72 | (0.40, 1.29) | 0.71 | (0.40, 1.26) | 0.72 | (0.40, 1.30) | 0.87 | (0.40, 1.92) |
| Other | 2.23 | (1.34, 3.70) | 2.3 | (1.37, 3.85) | 2.23 | (1.34, 3.72) | 2.45 | (1.43, 4.21) |
| Education | | | | | | | | |
| Degree or above | a | | a | | a | | a | |
| Certificate | 1.61 | (0.97, 2.65) | 1.47 | (0.89, 2.42) | 1.6 | (0.97, 2.65) | 1.89 | (0.99, 3.56) |
| School only | 1.59 | (0.98, 2.59) | 1.53 | (0.95, 2.45) | 1.58 | (0.97, 2.57) | 2.07 | (1.06, 4.06) |
| Less than 8th form | 2.31 | (1.10, 4.87) | 2.16 | (1.04, 4.5) | 2.31 | (1.10, 4.86) | 2.69 | (1.19, 6.11) |
| Undetermined | 0.33 | (0.03, 3.73) | 0.33 | (0.03, 3.69) | 0.33 | (0.03, 3.75) | 0.54 | (0.03, 10.9) |
| Number of LTC | | | | | | | | |
| Duration of care (years) | | | | | | | | |
| <1 | a | | a | | a | | a | |
| 1–9 | 0.54 | (0.31, 0.94) | 0.51 | (0.29, 0.89) | 0.53 | (0.30, 0.94) | 0.47 | (0.24, 0.93) |
| 10+ | 0.77 | (0.43, 1.37) | 0.73 | (0.41, 1.31) | 0.77 | (0.43, 1.37) | 0.66 | (0.29, 1.50) |
| Missing | 0.54 | (0.14, 2.08) | 0.59 | (0.15, 2.30) | 0.53 | (0.14, 2.07) | 0.47 | (0.10, 2.17) |
| Care assistance | | | | | | | | |
| No | a | | | | | | | |
| Yes | 0.91 | (0.64, 1.29) | 0.91 | (0.64, 1.30) | n.a. | | n.a. | |
| Dementia | | | | | | | | |
| No | | | | | | | a | |
| Yes | | | | | | | 1.89 | (1.01, 3.5) |
| Ancillary parameters | | | | | | | | |
| Constant term | 0.15 | (0.06, 0.36) | 0.16 | (0.07, 0.37) | 0.15 | (0.06, 0.34) | 0.11 | (0.03, 0.37) |
| <i>n</i> | 1,255 | | 1,255 | | 1,255 | | 890 | |

Notes: 95% CI: 95% confidence intervals; MESB: main English-speaking countries; OR: odds ratio; NILF: not in the labor force; Care assist.: care assistance.

^aComparison category.

(Mean VIF = 1.46) or omitted variable bias and the final specification is confirmed with the Hosmer–Lemeshow test (Model 1: $\chi^2 = 9.4$, $p < 0.1$; Model 2: $\chi^2 = 4.1$, $p < 0.1$). All models controlled for duration of care and prior use of care services.

Discussion

Like most high-income nations such as the USA and United Kingdom, Australia's population is projected to age considerably out to and beyond 2050 (ABS, 2013). The Australian government,

along with older persons themselves, prioritize care in the community, rather than institutional settings. This places not only considerable faith, but also pressure upon the informal care workforce in Australia, numbering in excess of 2.7 million people. Supporting the needs of carers is therefore crucial. In this study, we sought to (1) examine what are the unmet care needs of the primary carers of older Australians and (2) to investigate whether there is an association between these needs being unmet and the carers' mental health.

Types of unmet support needs

In 2015, 35% of carers of older Australians cited at least one unmet need for support. Among this group, almost two-thirds cited multiple unmet support needs (64.7%). The most prevalent types of unmet needs included financial (18%), physical (13%), emotional support (12%), as well as additional respite care (12%). These findings are consistent with earlier surveys of carers in Australia (Schofield *et al.*, 1998; Cummins *et al.*, 2007). The study by Cummins *et al.* (2007) found that, "satisfaction with ability to pay for household essentials, to afford the things you would like to have, to save money, to have financial security, and to not worry about income covering expenses, are all severely comprised for carers compared with a general population sample (p. vii)." They also found that carers were much less likely to be in paid employment compared with the general population with 20.6% of their sample classifying themselves as unemployed (Cummins *et al.*, 2007).

Our findings show that carers still have significant unmet financial needs. This could be related to the well-documented difficulties that carers have in combining paid work with caring, especially primary carers. It could also mean current carer payments are not adequate to meet the financial needs of those who rely on these payments. Future research could include a review of the effectiveness of current workplace arrangements that seek to enable carers to combine paid work with a caring role and/or a review of the adequacy of carer payments from government.

Other unmet needs we identify (physical and emotional support and additional respite care) are potentially due to a lack of services and/or poor tailoring of these services for the diversity of carer needs. Despite carers often identifying respite care as a need, they rarely take up the current respite offerings, as they are either inconvenient or difficult to access for the carer or do not provide suitable activities or care for the care recipient (Phillipson, 2016). Respite services should be better tailored to meet carers' and carer recipients' specific needs,

including offering culturally specific programs to carers from culturally and linguistically diverse background (Kosloski *et al.*, 2002).

It is interesting to note that those with multiple support needs were significantly more likely to signal needs for training and aids to assist in their caring role. There are existing free on-line training programs for carers of people with specific conditions usually associated with older age, such as dementia, in Australia. This study suggests that these could be better targeted to those carers who report a number of unmet needs.

Results here also underscore the importance of measurement in the unmet needs of carers. Focusing only on a single item measure of the "most assist you" significantly under represents the prevalence of a range of unmet needs of carers, which is important as two-thirds of those with unmet needs have multiple unmet support needs. As such, usage of the single item measure significantly distorts the types of overall needs of carers with multiple unmet needs, particularly as it pertains to needs for training and aids to assist in their caring role. This may have implications for health and age care professionals working to support carers. Careful wording of questions aimed to assess carers' unmet needs is required. Rather than asking a single question of carers as to what they think would be of most assistance, our findings suggest that a checklist with a range of options would enable carers to better identify their unmet needs.

Association between unmet needs and mental health

This measurement issue is noteworthy, as results of this study point to an important association between increasing unmet needs of carers and a higher likelihood of psychological distress. Accounting for complex survey design and incorporating extensive control variables, we find that having any unmet need for support increases the odds of psychological distress twofold and that each additional unmet need increased the likelihood of psychological distress by 36%. Given that two-thirds of carers in this sample have multiple unmet support needs, this result is concerning.

Carer depression has been found to be a risk factor for placement of the older care recipient in residential aged care (Gaugler *et al.*, 2009) and abuse of the care recipient (Cooper *et al.*, 2010), so it is a key risk factor for adverse outcomes in the care relationship. However, it is also important for the health and well-being of the carer themselves, as increased depression is associated with reduced quality of life (Bruvika *et al.*, 2012) and increased

suicidal ideation in carers (O'Dwyer *et al.*, 2013). Most of the research on caregiving has focused on characteristics of the carer, care recipient and the relationship between them rather than broader social and policy issues (Dow and McDonald, 2003). While a previous research has identified important directions for mental health clinicians (for example, Loi *et al.*, 2016), this study points to the need to consider broader policy issues, such as income support, as many carers clearly have unmet financial needs that may impact their mental health.

The availability of government programs, however, does not necessarily mean that carers take them up and/or that they reduce burden or mental distress. In Australia, less than 30% of carers use respite services (AIHW, 2007). There has been considerable investigation of why carers do not use respite services and the evidence is mixed. For example, some studies show spousal carers are less likely to use respite than non-spousal carers (Robinson *et al.*, 2005). Other studies have not found any relationship between carer relationship and use of respite services (Douglass and Visconti, 1998; Douglass and Fox, 1999; Montoro-Rodriguez *et al.*, 2003). Indeed, some studies show carer or caregiver characteristics are only weakly associated with non-use of services (Phillipson *et al.*, 2014). It may simply be that these services are difficult to access or do not meet carers' or care recipients' needs (Phillipson, 2016). For example, people from a culturally and linguistically diverse background are more likely to use services that are specifically tailored to their culture (Kosloski *et al.*, 2002).

Apart from barriers and usage to respite care, there have been very few studies in Australia on barriers and applications for financial support, for example, through the Carers Payment or Carers Allowance. This is a considerable research gap and further analysis is warranted here.

Limitations and extensions

In interpreting these results, it is important to recognize the studies limitations. First, the data are cross sectional. We cannot and do not draw causal inferences about carers' unmet needs and psychological distress. Unfortunately, there is a dearth of nationally representative longitudinal data with measures of carers' needs and psychological well-being in Australia, in which this linkage could be explored further.

An additional limitation of this study is that the SDAC measures focus only on primary carers. However, this group represents a substantial proportion of the total informal care workforce in

Australia. Of the 2.7 million Australians providing informal care in 2015, approximately 856,000, or about one-third, were deemed to be primary carers by the ABS (ABS, 2016b). Our study represents a sub sample of about 60% of this group who specifically care for older Australians. By definition, however, primary carers provide the most informal care to the recipient, and therefore most likely to have unmet needs to assist in their care provision. However, these data do raise the question of how other carers' unmet needs for support are distributed throughout the population and whether the associations with mental health are as strong as we observe in this study? For example, do carers of younger care recipients have differing unmet needs and does this association with psychological distress persist to the same degree observed here?

Analysis of longitudinal data to assess the relevant pathway from unmet support needs to psychological distress is a priority. Further studies may also wish to examine the relationship between unmet support needs and mental health in other countries with different funding and support mechanisms for carers and care recipients. Moreover, there is a need for further Australian studies on barriers to accessing means or alleviating financial stress – mainly through the Carers Allowance and Carers Payment. There is also considerable scope to examine the intersection of unmet needs, the care recipients' health conditions and carers' psychological well-being.

Summary

Notwithstanding these limitations, using newly released nationally representative data on Australian carers, we show the prevalence of unmet support needs to be considerable (34.6%) that two-thirds of this group have multiple unmet support needs and that unmet financial, emotional, physical, and support to improve carers' health are among the most commonly specified. Moreover, we show a clear association between unmet needs and psychological distress, independent of comprehensive control variables and accounting for complex survey design. We further show that even accounting for unmet needs, specific health conditions of the care recipient, such as dementia can heighten carers' likelihood of psychological distress.

Conflict of interest

None.

Description of authors' roles

J. B. Temple and B. Dow jointly designed the study and authored the manuscript. J. B. Temple performed the data analysis.

Acknowledgments

Data for this study were made available to the authors by the Australian Bureau of Statistics (ABS). J. B. Temple is funded by the Australian Research Council's Centre of Excellence in Population Ageing Research (CE1101029). The opinions expressed herein are those of the authors alone.

References

- Australian Bureau of Statistics (ABS)** (2013). *Catalogue Number 3222.0 – Population Projections, Australia, 2012 to 2101*. Canberra: Australian Bureau of Statistics.
- Australian Bureau of Statistics (ABS)** (2016a). *Catalogue Number 4430.0 – Disability, Ageing and Carers, Australia: Summary of Findings*. Canberra: Australian Bureau of Statistics.
- Australian Bureau of Statistics (ABS)** (2016b). *A Profile of Carers in Australia. Information Sheet*. Canberra: Australian Bureau of Statistics.
- Agincare** (2017). Available at: <https://www.agincare.com/articles/quit-job-to-care-for-parents-150227.htm>; last accessed August 2017.
- Anderson, T. M., Sunderland, M., Andrews, G., Titov, N., Dear, B. F. and Sachdev, P. S.** (2013). The 10-item Kessler psychological distress scale (K10) as a screening instrument in older individuals. *American Journal of Geriatric Psychiatry*, 21, 596.
- Australian Government** (2017). *Carer Gateway*. Available at: <https://www.carergateway.gov.au/>; last accessed August 2017.
- Australian Institute of Health and Welfare (AIHW)** (2007). *Dementia in Australia: National Data Analysis and Development*. Canberra: AIHW. Available at: <http://www.aihw.gov.au/publications/index.cfm/title/10368>.
- Belsley, D., Kuh, E. and Welsch, R.** (1980). *Regression Diagnostics: Identifying Influential Data and Sources of Collinearity*. New York: John Wiley & Sons, Inc.
- Bertrand, R., Fredman, L. and Saczynski, J.** (2006). Are all caregivers created equal? Stress in caregivers to adults with and without dementia. *Journal of Aging and Health*, 18, 534–551.
- Bruvika, F. K., Ulsteina, I. D., Ranhoff, A. H. and Engedala, K.** (2012). The quality of life of people with dementia and their family carers. *Dementia and Geriatric Cognitive Disorders*, 34, 7–14.
- Carers Australia** (2017). Available at: <http://www.carersaustralia.com.au/>, accessed August 2017.
- Carers UK** (2015). *Facts About Carers: Policy Briefing*. London: Carers UK.
- Cooper, C., Selwood, A., Blanchard, M., Walker, Z., Blizard, R. and Livingston, G.** (2010). The determinants of family carers' abusive behavior to people with dementia: results of the CARD study. *Journal of Affective Disorders*, 121, 136–142.
- Cummins, R. A., Hughes, J., Tomy, A., Gibson, A., Woerner, J. and Luffana, L.** (2007). *Special Report – The Wellbeing of Australians: Carer Health and Wellbeing*. Australian Unity Wellbeing Index, Survey 17.1, Report 17.1. Melbourne: Australian Unity.
- Dahlberg, L., Demack, S. and Bambra, C.** (2007). Age and gender of informal carers: a population-based study in the UK. *Health and Social Care in the Community*, 15, 439–445.
- Deloitte Access Economics** (2015). *The Economic Value of Informal Care in 2015*. Report for Carers Australia. Canberra: Deloitte Access Economics.
- Douglass, C. and Fox, P.** (1999). Health care utilization among clients with Alzheimer's disease: public policy implications from the California Alzheimer's Disease Diagnostic and Treatment Center Program. *Journal of Applied Gerontology*, 18, 99–121.
- Douglass, C. and Visconti, C.** (1998). Factors influencing the use of adult day care by individuals with Alzheimer's disease: a multivariate examination of the California Alzheimer's Disease Diagnostic and Treatment Center Program. *Home Health Care Services Quarterly*, 17, 53–69.
- Dow, B. and McDonald, J.** (2003). Social support or structural change? Social work theory and research on care giving. *Australian Social Work*, 56, 197–208.
- Dow, B., Sparrow, P., Moore, K., Gaffy, E. and Yates, I.** (2013). What do older Australians want? *Australasian Journal on Ageing*, 32, 236–240.
- Essue, B. et al.** (2010). Informal care and the self-management partnership: implications for Australian health policy and practice. *Australian Health Review*, 34, 414–422.
- Gaugler, J. E., Anderson, K. A., Leach, M. R., Smith, C. D., Schmitt, F. A. and Mendiondo, M.** (2004). The emotional ramifications of unmet need in dementia caregiving. *American Journal of Alzheimer's Disease and Other Dementias*, 19, 369–380.
- Gaugler, J. E., Yu, F., Krichbaum, K. and Wyman, J. F.** (2009). Predictors of nursing home admission for persons with dementia. *Medical Care*, 47, 191–198.
- Horner, B. and Boldy, D.** (2008). The benefit of 'ageing-in-place' in an aged care community. *Australian Health Review*, 32, 356–365.
- Hosmer, D., Hosmer, T., Le Cessie, S. and Lemeshow, S.** (1997). A comparison of goodness-of-fit tests for the logistic regression model. *Statistics in Medicine*, 16, 965–980.
- Kosloski, K., Schaefer, J., Allwardt, D., Montgomery, R. and Karner, T.** (2002). The role of cultural factors on clients' attitudes toward caregiving, perceptions of service delivery, and service utilization. *Home Health Care Services Quarterly*, 21, 65–88.
- Loi, S. M. et al.** (2016). Factors associated with depression in older carers. *International Journal of Geriatric Psychiatry*, 31, 294–301.

- Lucke, J. et al.** (2008). Few urban-rural differences in older carers' access to community services. *Australian Health Review*, 32, 684–690.
- Montoro-Rodriguez, J., Kosloski, K. and Montgomery, R.** (2003). Evaluating a practice-oriented service model to increase the use of respite services among minorities and rural caregivers. *The Gerontologist*, 43, 916.
- National Alliance for Caregiving** (2015). *Caregiving in the US*. Available at: http://www.caregiving.org/wp-content/uploads/2015/05/2015_CaregivingintheUS_Executive-Summary-June-4_WEB.pdf; last accessed 10 August 2017.
- Neri, A. L., Yassuda, M. S., Fortes-Burgos, A. C. and Guariento, M.** (2012). Relationships between gender, age, family conditions, physical and mental health, and social isolation of elderly caregivers. *International Psychogeriatrics*, 24, 472–483.
- O'Connell, B., Bailey, S. and Walker, A.** (2003). Promoting the health and well being of older carers: a proactive strategy. *Australian Health Review*, 26, 78–86.
- O'Dwyer, S., Moyle, W., Zimmer-Gembeck, M. and De Leo, D.** (2013). Suicidal ideation in family carers of people with dementia: a pilot study. *Geriatric Psychiatry*, 28, 1182–1188.
- Phillipson, L.** (2016). Rethinking dementia care. *Australian Journal of Dementia Care*. Available at: <http://journalofdementiacare.com/rethinking-respite-care/>; last accessed 24 November 2017.
- Phillipson, L., Jones, S. C. and Magee, C.** (2014). A review of the factors associated with the non-use of respite services by carers of people with dementia: implications for policy and practice. *Health and Social Care in the Community*, 22, 1–12.
- Productivity Commission** (2011). *Caring for Older Australians*. Report No. 53. Final Inquiry Report. Canberra: Productivity Commission.
- Raftery, A.** (1995). Bayesian model selection in social research. *Sociological Methodology*, 25, 111–163.
- Robinson, K., Buckwalter, K. and Reed, D.** (2005). Predictors of use of services among dementia caregivers. *Western Journal of Nursing Research*, 27, 126–140.
- Schofield, H., Bloch, S., Herrman, H., Murphy, B. and Nankervis, J.** (eds.) (1998). *Family Caregivers: Disability, Illness and Ageing*. Melbourne: Allen and Unwin.
- Stata Corporation** (2017). *Stata/SE 15.0 for Windows 64 Bit*. College Station, TX: Stata Corporation.
- Stirling, C., Andrews, S., Croft, T., Vickers, J., Turner, P. and Robinson, A.** (2010). Measuring dementia carers' unmet need for services – an exploratory mixed method study. *BMC Health Services Research*, 10, 122. doi:10.1186/1472-6963-10-122
- Vecchio, N.** (2008a). Understanding the use of respite services among informal carers. *Australian Health Review*, 32, 459–467.
- Vecchio, N.** (2008b). The use of support systems by informal caregivers: an Australian experience. *Australian Journal of Primary Health*, 14, 27–34.
- Victorian Equal Opportunity and Human Rights Commission** (2012). *Mature-Age Workers and the Equal Opportunity Act: Know Your Rights*. Melbourne: Victorian Equal Opportunity and Human Rights Commission.
- Winter, N.** (2008). SVR: Stata module to compute estimates with survey replication based standard errors. Available at: <https://ideas.repec.org/c/boc/bocode/s427502.html>; last accessed March 2017.
- Wolfer, K.** (1985). *Introduction to Variance Estimation*. New York: Springer-Verlag.

The effects of gender, age, schooling, and cultural background on the identification of facial emotions: a transcultural study

Leonardo Cruz de Souza,^{1,2,3} Maxime Bertoux,⁴ Ângelo Ribeiro Vaz de Faria,³
Laiane Tábata Souza Corgosinho,³ Ana Carolina de Almeida Prado,³
Izabela Guimarães Barbosa,^{1,3,5} Paulo Caramelli,^{1,2} Enrico Colosimo⁶
and Antônio Lúcio Teixeira^{1,2,3}

¹Programa de Pós-Graduação em Neurociências, Universidade Federal de Minas Gerais (UFMG), Belo Horizonte, MG, Brazil

²Departamento de Clínica Médica, Faculdade de Medicina da UFMG, Belo Horizonte, MG, Brazil

³Laboratório Interdisciplinar de Investigação Médica, Faculdade de Medicina da UFMG, Belo Horizonte, MG, Brazil

⁴Centre Mémoire de Ressources et de Recherche (CMRR), University Hospital of Lille (France) & Inserm U1171, Lille, France

⁵Departamento de Saúde Mental, Faculdade de Medicina da UFMG, Belo Horizonte, MG, Brazil

⁶Departamento de Estatística, UFMG, Belo Horizonte, MG, Brazil

ABSTRACT

Background: Social cognition tasks, such as identification of emotions, can contribute to the diagnosis of neuropsychiatric disorders. The wide use of Facial Emotion Recognition Test (FERT) is hampered by the absence of normative dataset and by the limited understanding of how demographic factors such as age, education, gender, and cultural background may influence the performance on the test.

Methods: We analyzed the influence of these variables in the performance in the FERT from the short version of the Social and Emotional Assessment. This task is composed by 35 pictures with 7 different emotions presented 5 times each. Cognitively healthy Brazilian participants ($n = 203$; 109 females and 94 males) underwent the FERT. We compared the performance of participants across gender, age, and educational subgroups. We also compared the performance of Brazilians with a group of French subjects ($n = 60$) matched for gender, age, and educational level.

Results: There was no gender difference regarding the performance on total score and in each emotion subscore in the Brazilian sample. We found a significant effect of aging and schooling on the performance on the FERT, with younger and more educated subjects having higher scores. Brazilian and French participants did not differ in the FERT and its subscores. Normative data for employing the FERT in Brazilian population is presented.

Conclusions: Data here provided may contribute to the interpretation of the results of FERT in different cultural contexts and highlight the common bias that should be corrected in the future tasks to be developed.

Key words: emotion, cognition, social cognition

Introduction

The assessment of cognitive dysfunction associated to neurological and psychiatric disorders is a clinical challenge, particularly in populations with heterogeneous educational background. The neuropsychological assessment is a critical step in the diagnostic investigation of neurodegenerative

diseases and psychiatric disorders, providing valuable markers for both diagnosis and follow-up of these patients. Recently, it has been recognized that social cognition tasks, such as identification of emotions, can critically contribute to the diagnosis of neuropsychiatric disorders (Buhl *et al.*, 2013, Kumfor and Piguet 2013). The American Psychiatric Association (2013) has consecutively included social cognition as one of the six main cognitive domains to evaluate in the new version of the Diagnostic and Statistical Manual of Mental Disorders (2013). Different cognitive batteries were proposed to assess social cognition in clinical practice (Torralva *et al.*, 2009; Bertoux *et al.*, 2012;

Correspondence should be addressed to: Leonardo Cruz de Souza, Faculdade de Medicina, Universidade Federal de Minas Gerais. Avenida Professor Alfredo Balena, no. 190/sl 243, Santa Efigênia, Belo Horizonte, MG CEP 30.130-100, Brazil. Phone number: + 55 31 3409 8073. E-mail: leocrusousa@hotmail.com. Received 7 May 2017; revision requested 14 Jan 2018; revised version received 25 Feb 2018; accepted 5 Mar 2018. First published online 25 May 2018.

Buhl *et al.*, 2013). Among them, the short version of the Social and Emotional Assessment (Mini-SEA) has been used to evaluate social cognition in neurological and psychiatric patients (Bertoux *et al.*, 2012). It consists of a theory of mind task (a modified version of the faux-pas test) and a Facial Emotion Recognition Test (FERT) composed by a selection of 35 faces from Ekman's pictures. It has been demonstrated that this battery provides accurate diagnostic differentiation between Alzheimer's disease and behavioral-variant frontotemporal dementia (bvFTD), and between bvFTD and major depressive disorder (Bertoux *et al.*, 2012).

One of the critical points concerning the wide use of Mini-SEA or other social cognition batteries in clinical practice is the absence of large normative dataset concerning the FERT. Indeed, most studies were limited by a restricted number of participants and by experimental design (Torralva *et al.*, 2009; Bertoux *et al.*, 2012; Buhl *et al.*, 2013). In the particular case of Mini-SEA, another critical issue is a better understanding of how demographic factors such as age, education, and gender, as well as cultural background, may influence the performance on the FERT. As a concrete example, it has been shown that there is an age-related decline in the recognition of facial emotions, independently from basic perception abilities (Ruffman *et al.*, 2008; Mill *et al.*, 2009; Grainger *et al.*, 2015). The effect of age on the performance on the Mini-SEA's FERT has not been specifically investigated in a large sample of subjects.

In addition, despite some innate aspects of emotion recognition ability, the cultural background and educational level may also modulate the emotion perception (Engelmann and Pogosyan, 2013). Similarly, the effect of schooling and cultural environment on the FERT has not been addressed. This is a crucial aspect for designing neuropsychological tests that could be used in multi-centric studies, which often include patients with different cultural backgrounds and heterogeneous educational level.

The aim of this study was to analyze the influence of gender, age, education, and cultural background in the FERT from the mini-SEA (Bertoux *et al.*, 2012). We, therefore, analyzed the effect of these demographics and cultural variables on the FERT's performance, in a large sample of Brazilian healthy subjects with heterogeneous educational level. The effect of cultural background on the same test was investigated by comparing the performance of Brazilian and French subjects matched for gender, age and educational level. Finally, we also provide normative data for the Brazilian population.

Methods

This study was conducted by the Cognitive and Behavioral Neurology Group of the School of Medicine from the Federal University of Minas Gerais (Belo Horizonte, Brazil). The study was approved by the Local Ethics Committee and subjects provided written informed consent before participating.

Participants

The Brazilian sample consisted of healthy controls recruited from the community on a voluntary basis. All participants underwent a standardized interview focused on neurological and psychiatric antecedents. Subjects were not included in the study if they presented any of the following criteria: (1) history of neurological disorder, such as stroke, epilepsy, multiple sclerosis, or neurodegenerative diseases; (2) history of neurosurgical procedures; (3) memory or cognitive complaints; (4) history of bipolar disorder or schizophrenia; (5) score above 8 out 21 either in anxiety or depression subscales from the Hospital Anxiety and Depression Scale (Botega *et al.*, 1995), and (6) subjects in use of medications that may interfere in cognitive performance (e.g. benzodiazepines). All participants were also evaluated by the Mini-Mental State Examination (MMSE) (Nitrini and Caramelli, 2003), and we only included individuals who scored above norms adjusted for educational level. The following cut-offs were considered for the Brazilian participants: higher than or equal to 20 for illiterates, higher than or equal to 21 (for individuals with one to three years of education), higher than or equal to 24 (for subjects with four to seven years of education), and higher than or equal to 26 (for subjects with eight or more years of education) (Nitrini and Caramelli, 2003).

We stratified the Brazilian population into seven groups according to age as follows: Group 1 (15–20 years); Group 2 (21–30 years); Group 3 (31–40 years); Group 4 (41–50 years); Group 5 (51–60 years); Group 6 (61–70 years) and Group 7 (≥ 70 years).

The Brazilian sample was also stratified into four groups according to years of education as follows: Group A consisting of illiterate participants; Group B consisting of participants with 1–3 years of formal schooling; Group C formed by the participants with 4–7 years of formal schooling; Group D formed by the participants with 8–11 years of formal schooling; and Group E formed by the participants who had 12 or more years of formal schooling. Illiteracy was here defined as the inability to read the phrase "Close your eyes" from the MMSE.

In order to investigate the possible effect of cultural background on the performance on the FERT, we compared a subgroup of Brazilian participants ($n = 60$) with 60 healthy French subjects (Table 4), from a previous study (Bertoux *et al.*, 2015). We selected 60 Brazilian subjects from the full sample ($n = 203$), individually matching them with French participants, for gender, age (± 2 years), and educational level (± 2 years). This procedure was adopted to avoid a selection bias caused by these variables. French participants were selected under similar inclusion criteria used in the Brazilian sample: no cognitive complaint, absence of neurological, neurosurgical or psychiatric history, and normal score on the MMSE.

Identification of emotions

We used the FERT included in the mini-SEA (Bertoux *et al.*, 2015). This task is composed by a selection of 35 pictures from Ekman's portfolio (Ekman and Friesen, 1975), which are presented in a screen for 12 s. Seven different emotions (happiness, sadness, fear, disgust, surprise, anger, and neutral) are presented five times each, in a pseudo-randomized order. Male and female faces are presented in a similar ratio. Labels of emotions are presented at the same time on the screen during the entire task, so that the performance is independent of memory processing. Participants orally indicated which emotion was expressed and their answers were then registered.

Statistical analyses

All statistical analyses were performed using the Statistical Package for Social Sciences (SPSS version 22) and R Software (2016). Descriptive statistics were performed to characterize the sample and each group of participants. Normality assumption was verified by the Kolmogorov–Smirnov test and also by histograms visual inspection. Non-parametric tests were used in the data analysis, since there was evidence of non-normality.

χ^2 test was used for comparing categorical (gender) variable among groups. Jonckheere–Terpstra test was employed for comparing non-parametric continuous variables among Brazilian subgroups according to age and educational level, and, when pertinent, Mann–Whitney U test (with Bonferroni correction) was applied to perform two-by-two group comparisons. Mann–Whitney U test was also employed to compare gender groups (male vs. female). We used Wilcoxon sign rank test to compare Brazilian and French groups on the FERT.

Local smooth technique, Lowess (Cleveland, 1993), was used to describe the behavior of total score (TS) and age and schooling. Lowess is a data-driven technique. Non-linear regression was fitted for the data based on a linear spline with one knot for age and another one for schooling. This model was necessary to obtain the equivalent score that is described elsewhere (Dodich *et al.*, 2014). Equivalent score make possible direct comparison among participants. We also carried out analysis with Spearman's test to investigate correlations between FERT (TS) and age. A similar approach was adopted to explore correlations between FERT and educational level.

Results

The final sample of study consisted of 203 Brazilian participants (109 females and 94 males) with mean age 48.8 ± 19 years (range 15–86 years) and mean educational level of 9.3 ± 5.1 years (range 0–22 years). Table 1 presents socio-demographic and clinical data for Brazilian sample.

Effect of gender

Age and schooling were similar between men and women. There was no gender difference regarding the performance on the TS and in each emotion from the FERT in the Brazilian sample (Table 1). Women performed better than men on the TS in the French group, without any specific difference in each emotion.

Effect of age

Jonckheere–Terpstra test showed differences across groups stratified according to age, in the TS ($p < 0.001$) and in each category (Surprise: $p < 0.001$ / Fear: $p < 0.001$ / Anger: $p < 0.001$ / Neutral: $p < 0.01$), except for happiness, disgust, and sadness ($p > 0.05$ for all). Then, two-by-two group comparisons were conducted with Mann–Whitney test, with Bonferroni correction (Table 2).

Briefly, younger subjects performed better than older ones. Group 1 (15–20 years) did not differ from Group 2 (21–30 years), 3 (31–40 years), and 4 (41–50 years). There was difference between Groups 1 and 5 (51–60 years) in TS and Surprise. There was a trend of Group 1 to perform better than Groups 6 (61–70 years) and 7 (older than 70 years) in TS, Surprise, and Neutral subscores, but these differences did not survive after Bonferroni correction. Comparisons between other groups are presented in Table 2. The performance on the FERT decreases progressively until by 50 years, and then stabilizes (Figure

Table 1. Demographic and neuropsychological data for the Brazilian sample

| | TOTAL SAMPLE (<i>N</i> = 203) MEDIAN ± SD (P25–P75) | FEMALE (<i>N</i> = 109) MEDIAN ± SD (P25–P75) | MALE (<i>N</i> = 94) MEDIAN ± SD (P25–P75) | P (MANN– WHITNEY TEST) (FEMALE VS. MALE) |
|-----------------------------------|---|---|--|---|
| Age (years) | 50 ± 19.2 (31–66) | 54 ± 19.4 (31–67) | 46 ± 18.9 (31–65) | 0.36 |
| Schooling (years of education) | 10 ± 5.1 (5–14) | 10 ± 4.9 (5–13) | 10 ± 4.51 (5–14) | 0.75 |
| MMSE (/30) | 28 ± 2.7 (26–29) | 28 ± 2.8 (26–29) | 28 ± 2.8 (26–29) | 0.42 |
| FERT – total score (/35) | 27 ± 4.4 (23–30) | 27 ± 4.4 (23–29) | 27 ± 4.5 (23–33) | 0.76 |
| FERT – happiness (/5) | 5 ± 0.4 (5–5) | 5 ± 0.3 (5–5) | 5 ± 0.26 (5–5) | 0.66 |
| FERT – surprise (/5) | 4 ± 1.1 (3–5) | 4 ± 1.1 (3–5) | 4 ± 1.07 (3–5) | 0.72 |
| FERT – disgust (/5) | 4 ± 1.1 (3–5) | 4 ± 1.1 (3–5) | 4 ± 0.89 (3–5) | 0.84 |
| FERT – fear (/5) | 2 ± 1.3 (2–4) | 2 ± 1.2 (2–3) | 2 ± 1.44 (2–4) | 0.98 |
| FERT – anger (/5) | 3 ± 1.1 (3–4) | 4 ± 1.1 (3–4) | 3 ± 1.09 (3–4) | 0.77 |
| FERT – sadness (/5) | 4 ± 1.1 (3–5) | 4 ± 1.1 (3–5) | 4 ± 1.05 (3–5) | 0.99 |
| FERT – neutral (/5) | 5 ± 1.0 (4–5) | 5 ± 1.1 (3–5) | 5 ± 0.93 (4–5) | 0.29 |

FERT = Facial Emotion Recognition Test; P25 = 25th percentile; P75 = 75th percentile; MMSE = Mini-Mental State Examination; SD = Standard deviation. The significance of *p* was set at 0.0045 (Bonferroni correction).

S1, available as supplementary material attached to the electronic version of this paper at www.journals.cambridge.org/jid_IPG). There was a significant negative correlation (Spearman's correlation test) between TS (FERT) and age ($p < 0.001$, $r = -0.38$).

Effect of education

The third objective of this study was to analyze the influence of educational level on the FERT. Jonckheere–Terpstra test showed differences related to schooling subgroups in the TS and in each emotion category in the test. Illiterates (Group A) performed worse than all other groups, except Group B (1–3 years of education). Two-by-two group comparisons found no significant differences between groups B (1–3 years) and C (4–7 years) in TS and all Ekman Faces Test categories (Table 3). Although we found a significant value for Fear score, it was not significant after Bonferroni correction. Comparing Group B (1–3 years) with D (8–11 years), we noticed significant differences in the TS and in all Ekman Faces Test categories, except for Fear and Happiness score. The comparison between groups B (1–3 years) and E (12 or more years) showed differences in the TS and in all FERT subscores, except for Fear. Comparing Group C (4–7 years) with D (8–11 years), we noticed significant differences in all FERT scores, except for Happiness. The comparison between Group C (4–7 years) and E (12 or more years) revealed differences in all FERT scores. Comparing groups D (8–11 years) and E (12 or more years),

we observed no significant differences. In sum, groups with higher educational level performed better than participants with lower education. The performance on the FERT increases progressively until by 11 years of education, and then stabilizes (Figure S1B). There was a significant positive correlation (Spearman's correlation test) between TS (FERT) and educational level ($p < 0.001$, $r = 0.70$).

Effect of cultural background

The fourth objective of the present study was to assess the effect of different cultural backgrounds on the performance on the identification of emotions. We adopted a transcultural approach, by comparing two groups of participants (Brazilians vs. French), matched for gender, age, and educational level (Table 4). When comparing Brazilians and French (Wilcoxon sign rank test), we found no significant differences in TS and in each emotion. There was a trend for Brazilian scoring lower than French in surprise and disgust, but these differences did not remain significant after Bonferroni correction.

Normative data for the Brazilian population

Table 5 provides normative data for the Brazilian population, considering age and education as predictors of performance. Briefly, TS is positively correlated with schooling and inversely with age. Equivalent score is obtained after removing age and schooling effect by using the fitted non-linear regression.

Table 2. Demographic and neuropsychological data across age groups in the Brazilian sample

| | GROUP 01 (15-20 YEARS) MEDIAN \pm SD (P25-P75) | GROUP 02 (21-30 YEARS) MEDIAN \pm SD (P25-P75) | GROUP 03 (31-40 YEARS) MEDIAN \pm SD (P25-P75) | GROUP 04 (41-50 YEARS) MEDIAN \pm SD (P25-P75) | GROUP 05 (51-60 YEARS) MEDIAN \pm SD (P25-P75) | GROUP 06 (61-70 YEARS) MEDIAN \pm SD (P25-P75) | GROUP 07 (>70 YEARS) MEDIAN \pm SD (P25-P75) |
|--------------------------------|---|---|---|---|---|---|---|
| Number | 12 | 37 | 29 | 24 | 33 | 39 | 29 |
| Age (years) | 18 \pm 1.5 (17-19) | 24 \pm 3.0 (22-27) ^{c,d,e} | 36 \pm 3.1 (33-39) ^{f,g,h} | 47 \pm 2.7 (44-48) ⁱ | 55 \pm 2.5 (55-58) | 67 \pm 2.9 (65-69) | 75 \pm 5.2 (73-79) |
| Schooling (Years of education) | 12 \pm 1.0 (10-12) | 14 \pm 3.1 (11-15) ^{c,d,e} | 11 \pm 4.4 (8-13) | 9 \pm 4.2 (5-11) | 7 \pm 6.0 (2-11) | 8 \pm 5.0 (5-12) | 5 \pm 5.2 (3-11) |
| MMSE (/30) | 30 \pm 2.7 (28-30) ^{a,b} | 28 \pm 1.2 (28-29) ^{c,e} | 28 \pm 1.9 (27-29) | 28 \pm 2.6 (25-29) | 27 \pm 3.3 (25-29) | 28 \pm 2.6 (26-29) | 27 \pm 3.3 (24-28) |
| FERT - total score (/35) | 28 \pm 3.3 (27-31) ^a | 29 \pm 2.0 (28-31) ^{d,e} | 28 \pm 2.8 (26-29) ^f | 27 \pm 4.7 (23-31) | 23 \pm 4.8 (20-28) | 26 \pm 4.8 (22-28) | 24 \pm 4.5 (21-29) |
| FERT - happiness (/5) | 5 \pm 0 (5-5) | 5 \pm 0.2 (5-5) | 5 \pm 0.4 (5-5) | 5 \pm 0.3 (5-5) | 5 \pm 0.5 (5-5) | 5 \pm 0.3 (5-5) | 5 \pm 0.5 (5-5) |
| FERT - surprise (/5) | 5 \pm 0.7 (3-5) ^a | 5 \pm 0.8 (4-5) ^{c,d,e} | 4 \pm 0.9 (4-5) ^f | 5 \pm 0.8 (4-5) ⁱ | 3 \pm 1.3 (2-4) | 4 \pm 1.1 (3-5) | 4 \pm 1.2 (3-5) |
| FERT - disgust (/5) | 4 \pm 0.7 (4-4) | 4 \pm 0.8 (4-5) | 4 \pm 1.0 (3-5) | 4 \pm 1.1 (3-5) | 3 \pm 1.2 (3-4) | 4 \pm 1.3 (3-5) | 4 \pm 1.0 (3-5) |
| FERT - fear (/5) | 3 \pm 1.5 (2-4) | 3 \pm 1.1 (2-4) ^c | 3 \pm 1.2 (2-4) ^{f,g,h} | 3 \pm 1.5 (2-4) | 2 \pm 1.2 (1-2) | 2 \pm 1.3 (1-3) | 2 \pm 1.1 (1-3) |
| FERT - anger (/5) | 4 \pm 1.0 (3-4) | 4 \pm 0.7 (3-4) ^{c,d,e} | 4 \pm 1.0 (3-4) ^h | 4 \pm 0.9 (3-4) | 3 \pm 1.3 (2-4) | 3 \pm 1.0 (3-4) | 3 \pm 1.0 (2-4) |
| FERT - sadness (/5) | 4 \pm 1.2 (3-4) | 4 \pm 0.9 (4-5) ^c | 4 \pm 0.9 (3-4) | 4 \pm 1.2 (3-5) | 3 \pm 1.3 (2-4) | 4 \pm 1.1 (3-5) | 3 \pm 1.1 (3-4) |
| FERT - neutral (/5) | 5 \pm 0.4 (5-5) | 5 \pm 0.5 (4-5) ^{c,d,e} | 4 \pm 0.9 (4-5) | 5 \pm 1.1 (3-5) | 4 \pm 1.2 (3-5) | 4 \pm 1.3 (3-5) | 4 \pm 1.0 (3-5) |

FERT = Facial Emotion Recognition Test; P25 = 25th percentile; P75 = 75th percentile; MMSE = Mini-Mental State Examination; SD = Standard deviation

There was not any difference between Groups 1 and 2, Groups 1 and 3, Groups 1 and 4, Groups 1 and 6, Groups 1 and 7, for all FERT scores.

There was not any difference between Groups 2 and 3, Groups 2 and 4, for all FERT scores.

There was not any difference between Groups 3 and 4, Groups 2 and 4, Groups 2 and 5, for all FERT scores.

There was not any difference between Groups 4 and 6, Groups 4 and 7, for all FERT scores.

There was not any difference between Groups 5 and 6, Groups 5 and 7, for all FERT scores.

There was not any difference between Groups 6 and 7, for all FERT scores.

^ap < 0.0045 (Mann-Whitney test, Bonferroni correction): Group 1 versus Group 5.

^bp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group 1 versus Group 7.

^cp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group 2 versus Group 5.

^dp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group 2 versus Group 6.

^ep < 0.0045 (Mann-Whitney test, Bonferroni correction): Group 2 versus Group 7.

^fp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group 3 versus Group 5.

^gp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group 3 versus Group 6.

^hp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group 3 versus Group 7.

ⁱp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group 4 versus Group 5.

Table 3. Demographic and Neuropsychological data across schooling groups in the Brazilian sample

| | GROUP A ILLITERATES MEDIAN ± SD (P25-P75) | GROUP B (1-3 YEARS OF EDUCATION) MEDIAN ± SD (P25-P75) | GROUP C (4-7 YEARS OF EDUCATION) MEDIAN ± SD (P25-P75) | GROUP D (8-11 YEARS OF EDUCATION) MEDIAN ± SD (P25-P75) | GROUP E (≥ 12 YEARS OF EDUCATION) MEDIAN ± SD (P25-P75) |
|--------------------------|--|--|--|---|---|
| Number | 14 | 18 | 44 | 60 | 68 |
| Age (years) | 68 ± 13.9 (56-74) ^{b,c} | 56 ± 9.2 (51-60) ^c | 60 ± 17.1 (44-68) ^g | 60 ± 17.1 (44-68) | 33 ± 19.5 (23-57) |
| MMSE (/30) | 20 ± 2.4 (19-21) ^{a,c} | 24 ± 2.3 (22-25) ^{d,e} | 26 ± 1.6 (25-27) ^{f,g} | 28 ± 1.1 (26-30) | 29 ± 1.1 (28-29) |
| FERT - Total score (/35) | 19 ± 3.4 (17-20) ^{a,b,c} | 22 ± 5.2 (18-26) ^{d,e} | 23 ± 2.5 (22-25) ^{f,g} | 28 ± 2.7 (27-30) | 30 ± 2.3 (28-31) |
| FERT - Happiness (/5) | 4 ± 0.7 (4-5) ^{a,b,c} | 5 ± 0.4 (5-5) ^e | 5 ± 0.4 (5-5) ^g | 5 ± 0.2 (5-5) | 5 ± 0.1 (5-5) |
| FERT - Surprise (/5) | 3 ± 0.9 (2-3) ^{b,c} | 3 ± 1.6 (2-5) ^{d,e} | 3 ± 1.1 (3-4) ^{f,g} | 5 ± 0.8 (4-5) | 5 ± 0.8 (4-5) |
| FERT - Disgust (/5) | 2 ± 1.0 (2-3) ^{a,b,c} | 3 ± 1.2 (3-4) ^{d,e} | 3 ± 0.9 (3-4) ^{f,g} | 4 ± 1.0 (4-5) | 4 ± 0.9 (4-5) |
| FERT - Fear (/5) | 2 ± 0.8 (1-2) ^c | 2 ± 1.1 (2-3) | 2 ± 1.1 (1-2) ^{f,g} | 3 ± 1.4 (2-4) | 3 ± 1.3 (2-4) |
| FERT - Anger (/5) | 2 ± 0.8 (2-3) ^{b,c} | 3 ± 1.2 (2-3) ^{d,e} | 3 ± 0.8 (3-3) ^{f,g} | 4 ± 0.9 (3-4) | 4 ± 0.9 (3-5) |
| FERT - Sadness (/5) | 3 ± 1.2 (2-3) ^{b,c} | 3 ± 1.2 (2-4) ^{d,e} | 3 ± 0.8 (3-3) ^{f,g} | 4 ± 1.0 (3-5) | 4 ± 0.8 (4-5) |
| FERT - Neutral (/5) | 4 ± 0.9 (3-4) ^{b,c} | 3 ± 1.5 (2-4) ^{d,e} | 3 ± 1.1 (3-4) ^{f,g} | 5 ± 0.6 (4-5) | 5 ± 0.4 (5-5) |

FERT = Facial Emotion Recognition Test; P25 = 25th percentile; P75 = 75th percentile; MMSE = Mini-Mental State Examination; SD = Standard deviation
 There was not any difference between Groups A and B, for all FERT scores.

There was not any difference between Groups B and C, for all FERT scores.

There was not any difference between Groups D and E, for all FERT scores.

^ap < 0.0045 (Mann-Whitney test, Bonferroni correction): Group A versus Group C.

^bp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group A versus Group D.

^cp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group A versus Group E.

^dp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group B versus Group D.

^ep < 0.0045 (Mann-Whitney test, Bonferroni correction): Group B versus Group E.

^fp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group C versus Group D.

^gp < 0.0045 (Mann-Whitney test, Bonferroni correction): Group C versus Group E.

Table 4. Demographic and Neuropsychological data for groups according to cultural backgrounds (France and Brazil)

| | FRENCH SAMPLE (N = 60) MEDIAN ± SD (P25–P75) | BRAZILIAN SAMPLE (N = 60) MEDIAN ± SD (P25–P75) | P (WILCOXON SIGN RANK TEST) |
|-------------------------------------|--|--|--------------------------------|
| Male/female | 28/32 | 28/32 | X |
| Age (years) | 50.5 ± 15.6 (42–68) | 49.5 ± 15.1 (39–66) | 0.81 |
| Schooling (years of education) | 12.0 ± 3.3 (9–15) | 11.0 ± 2.5 (10–14) | 0.78 |
| Mini-Mental State Examination (/30) | 29 ± 0.9 (28–30) | 29 ± 1.1 (28–29) | 0.05 |
| FERT – total score (/35) | 29 ± 2.6 (27–31) | 29 ± 2.8 (22–34) | 0.42 |
| FERT – happiness (/5) | 5.0 ± 0.2 (5–5) | 5.0 ± 0 (5–5) | 0.16 |
| FERT – surprise (/5) | 5.0 ± 0.6 (4–5) | 4.0 ± 0.7 (4–5) | 0.02 |
| FERT – Disgust (/5) | 5.0 ± 0.8 (4–5) | 4.0 ± 1.0 (3–5) | 0.02 |
| FERT – fear (/5) | 3.0 ± 1.2 (2–4) | 3.0 ± 1.4 (2–4) | 0.99 |
| FERT – anger (/5) | 4.0 ± 0.9 (3–4) | 4.0 ± 0.9 (3–5) | 0.98 |
| FERT – sadness (/5) | 4.0 ± 1.16 (3–4) | 4.0 ± 0.9 (3–5) | 0.12 |
| FERT – neutral (/5) | 5.0 ± 0.5 (5–5) | 5.0 ± 0.6 (5–5) | 0.49 |

FERT = Facial Emotion Recognition test; P25 = 25th percentile; P75 = 75th percentile; MMSE = Mini-Mental State Examination; SD = Standard deviation. The significance of p was set at 0.0045 (Bonferroni correction).

Table 5. Normative data for Brazilian population, with age and education corrections for the Facial Emotion Recognition Test (FERT)

| | AGE (YEARS) | | | | | | | | | | | | | |
|--------------------------------------|-------------|-------|-------|-------|-------|-------|-------|-------|-------|-------|-------|-------|-------|-------|
| | 15 | 20 | 25 | 30 | 35 | 40 | 45 | 50 | 55 | 60 | 65 | 70 | 75 | 80 |
| Education (years of schooling) | | | | | | | | | | | | | | |
| 0 | 5.92 | 6.15 | 6.37 | 6.60 | 6.83 | 7.06 | 7.29 | 7.52 | 7.53 | 7.55 | 7.56 | 7.58 | 7.59 | 7.61 |
| 4 | 2.43 | 2.66 | 2.88 | 3.11 | 3.34 | 3.57 | 3.80 | 4.02 | 4.04 | 4.05 | 4.07 | 4.08 | 4.10 | 4.11 |
| 8 | –1.06 | –0.84 | –0.61 | –0.38 | –0.15 | 0.08 | 0.31 | 0.53 | 0.55 | 0.56 | 0.58 | 0.59 | 0.61 | 0.63 |
| 11 | –3.68 | –3.45 | –3.23 | –3.00 | –2.77 | –2.54 | –2.31 | –2.08 | –2.07 | –2.05 | –2.04 | –2.02 | –2.01 | –1.98 |
| 15 | –3.82 | –3.59 | –3.36 | –3.14 | –2.91 | –2.68 | –2.45 | –2.22 | –2.21 | –2.19 | –2.18 | –2.16 | –2.15 | –2.12 |

Corrections for FERT score:

1. If age <50 years and education <11 years of schooling: Corrected score = Raw Score + 0.046 × (age–48.80)–0.873 (education–9.27)–0.624.
2. If age <50 years and education > 11 years of schooling: Corrected score = Raw Score + 0.046 × (age–48.80) –0.035 (education–9.27) –2.060.
3. If age > 50 years and education < 11 years of schooling: Corrected score = Raw Score + 0.003 × (age–48.80) –0.873 (education–9.27) –0.573.
4. If age > 50 years and education > 11 years of schooling: Corrected score = Raw Score + 0.003 × (age–48.80) –0.035 (education–9.27) –2.008.
5. The corrected scores can be classified into categories (Equivalent scores): 0 (0–20.97), 1 (20.98–22.76), 2 (22.77–24.55), and 3 (> 24.55). Equivalent score 3 refers to the values equal to or higher than median; 1 and 2 are intermediate intervals; and 0 refers to values below tolerance limit.

Discussion

This study analyzed the effect of gender, age, education, and cultural background on the performance on the FERT from the mini-SEA. We found a significant effect of aging and schooling on the performance on the FERT, with younger and more educated subjects having higher scores, in agreement with previous studies (Ruffman *et al.*, 2008; Engelmann and Pogosyan 2013; Dodich *et al.*, 2014).

We did not find any gender difference on any scores of the FERT in the Brazilian group; however, in the French sample, women performed better than men on the TS. Previous studies have shown mixed findings on gender difference in emotion recognition tasks, with some works reporting no gender differences (Derntl *et al.*, 2010; Khawar *et al.*, 2013) and others reporting that female participants tend to perform better than male (Demenescu *et al.*, 2014; Dodich *et al.*, 2014; Lawrence *et al.*, 2015). An interesting

point is that this difference was found in the French sample only, and not in the total Brazilian sample. Unfortunately, this study does not have the data to go beyond this result and this aspect should be studied in a more specific way in further studies, as we believe cultural gender specific factors could play an important role in social cognitive assessment, such as emotion recognition. By investigating the possible links between gender variation of emotion perception and measures of gender equality and perceived stereotypes through a transcultural approach, we believe that these further works could have a critical interest to interpret this reported difference and enhance our understanding of this gender effect.

We found a negative correlation between age and performance on the FERT (Figure S1A). Previous studies demonstrated age-related decline in different cognitive domains, especially in executive functions such as speed processing. This has been interpreted as a decrease of fluid intelligence, a factorial component of general cognitive abilities that is highly dependent on frontal regions (Roca *et al.*, 2013). Whether social cognition and emotion recognition in particular relies on fluid intelligence remains an open question. However, it has been recently shown that social cognition abilities are decreased in elderly in comparison with young participants (Ruffman *et al.*, 2008; Halberstadt *et al.*, 2011; Grainger *et al.*, 2015). This age-related decline seems independent from perceptual factors and would be related to cognitive processing involved in social abilities that are more dependent of fronto-temporal circuits or the so-called “social brain” (Ruffman *et al.*, 2008). Using neuro-imaging approaches, previous works did indeed show that normal, age-related, structural, and metabolic alterations take place in the brain, with the frontal cortex being the region where the greatest deterioration was observed (Kalpouzos *et al.*, 2009). These age differences in cognitive performance and frontal integrity should be accounted in the assessment of social cognition abilities in clinical settings and highlight the need of specific age category normative data in order to support clinical decisions based on this test.

Schooling also seems to modulate the accuracy of facial recognition. We found that more educated subjects performed better than less educated participants, in agreement with previous observations in Italian population (Dodich *et al.*, 2014). We also found significant positive correlations between years of schooling and FERT scores. Education plays a major role in modulating the performance of different cognitive abilities, such as executive

functions, language, and episodic memory (Nitrini *et al.*, 2004; Opdebeeck *et al.*, 2016). While the role of education could appear as being less obvious in emotion recognition than in other cognitive functions such as language, it is important to specify that the FERT relies on verbal labeling of emotions, as most of emotion recognition tasks. Recently, Lindquist *et al.* (2014) have shown evidences that identifying the emotion’s appropriate category (e.g. disgust, sadness, and surprise) depends heavily on the preliminary knowledge of the emotional concepts, independently from affective or implicit valence processing that first determine if an emotion is positive or negative (Lindquist *et al.*, 2014). It is possible that this specific semantic aspect of emotion recognition, which could be described as a taxonomy of emotion concepts, could be modulated by the level of education, as opposed to a more culture-free affective processing of the valence.

Brazilian and French participants did not differ in the FERT and its subscores for each emotion. Cross-cultural comparisons of population in emotion recognition abilities have been extensively investigated in the last decades (Ekman *et al.*, 1969, 1987; Russell, 1994; Engelmann and Pogosyan, 2013). These studies provided data that demonstrated the universality of facial expression of emotions, thus replacing the preceding thesis that facial emotions are not universal, but a cultural contingency. Indeed, there is a large scope of experimental and observational data showing a high accuracy among different cultures in the identification of basic emotions. For instance, studies comparing western versus non-western populations and also illiterate versus literate support the thesis of the universality of facial expressions (Russell, 1994). Of note, data from the seminal observations of Ekman and collaborators in pre-literate isolated societies in New Guinea and Borneo supported the thesis of a similar pattern of facial muscular activation for expression of basic emotions across cultures (Ekman *et al.*, 1969). In sum, these data argue for a common neurobiological basis for emotional processing, which would have a similar evolutionary origin (Ekman *et al.*, 1987; Engelmann and Pogosyan, 2013). Despite these common bases, there are cultural factors that modulate the facial expression, such as the “display rules.” These are social codes that regulate masking, inhibiting, or amplifying the expression of facial emotions in a given social context that critically depend on culture differences (Russell, 1994; Engelmann and Pogosyan 2013). There are also cultural determinants of factors that elicit emotions (Russell, 1994). These factors and methodological issues (e.g. tasks with posed

or spontaneous expressions of facial emotions) may account for some differences observed in cross-cultural studies. For instance, differences in overall recognition scores were described between Canadians and Ethiopians, Americans and Zambians, and Americans and Japanese (Russell, 1994; Engelmann and Pogosyan 2013). In the current study, we did not find differences between French and Brazilians in emotion recognition, differently from a previous study (Russell, 1994), which found that, compared to French subjects, Brazilians scored lower in the recognition of surprise, anger, and fear; and better in happiness, sadness, and disgust identification. The reasons for this difference remain unclear.

Here, we provide normative data for employing the FERT in the Brazilian population. This is of utmost importance due to the lack of clinical tools to assess social cognition in clinical practice.

In sum, this study highlights the effects of age and education in the accurate recognition of basic emotions, in agreement with previous studies. However, it was not possible to isolate the specific effect of age or schooling on the FERT, independently from each other. The educational level across age groups was not homogeneous, as the oldest group has lower educational level than the youngest. Considering Brazilian social context, this is due to the fact that the oldest group was of school age when schooling was not accessible to most of population. On the other hand, most of young people have been to school on the last three decades. More studies are necessary to understand the relationship of age and education on the FERT.

These data raise the question whether it is appropriate or valid to use tests of emotion recognition in populations with heterogeneous educational level and distinct cultural backgrounds. In the light of multicenter studies designed for including patients from different countries, there is an urge for developing neuropsychological tools that may be employed in different cultural settings, including populations with low educational level. This is very relevant as there is an increase in the number of demented patients in developing countries, with a majority of low educated patients among them (Nitrini *et al.*, 2009). Data provided by the present study may contribute for the interpretation of the results of FERT in different cultural contexts and highlight the common bias that should be corrected in future tasks to be developed. In that perspective, these data may help in the design of new tools for evaluating emotion recognition and other social abilities.

Conflict of interest

None.

Description of authors' roles

LCS designed the study, collected data, performed statistical analyses, and drafted the first version of the manuscript. MB designed the study, collected data, and critically reviewed the manuscript for intellectual content. ARVF and LTSC collected data and critically reviewed the manuscript for intellectual content. ACAP, IGB, and EC performed statistical analyses and critically reviewed the manuscript for intellectual content. PC critically reviewed the manuscript for intellectual content. ALT designed the study and critically reviewed the manuscript for intellectual content.

Acknowledgment

This study was partially funded by CNPq (402853/2012-1)

Supplementary material

To view supplementary material for this article, please visit <https://doi.org/10.1017/S1041610218000443>

References

- American Psychiatric Association** (2013). *Diagnostic and Statistical Manual of Mental Disorders (DSM-V)*. Washington, DC: American Psychiatric Association.
- Bertoux, M., de Souza, L. C., Sarazin, M., Funkiewiez, A., Dubois, B. and Hornberger, M.** (2015). How preserved is emotion recognition in Alzheimer disease compared with behavioral variant frontotemporal dementia?. *Alzheimer Disease and Associated Disorders*, 29, 154–7.
- Bertoux, M., . . . et al.** (2012). Social cognition and emotional assessment differentiates frontotemporal dementia from depression. *The Journal of Neurology, Neurosurgery, and Psychiatry* 83, 411–6.
- Botega, N. J., Bio, M. R., Zomignani, M. A., Garcia, C. Jr. and Pereira, W. A.** (1995). Mood disorders among inpatients in ambulatory and validation of the anxiety and depression scale HAD. *Revista de Saude Publica*, 29, 355–63.
- Buhl, C., Stokholm, J. and Gade, A.** (2013). Clinical utility of short social cognitive tests in early differentiation of behavioral variant frontotemporal dementia from Alzheimer's disease. *Dementia and Geriatric Cognitive Disorders Extra*, 3, 376–85.

- Cleveland, W. S.** (1993). *Visualizing Data*. Summit, NJ: Hobart Press.
- Demencescu, L. R., Mathiak, K. A. and Mathiak, K.** (2014). Age- and gender-related variations of emotion recognition in pseudowords and faces. *Experimental Aging Research*, 40, 187–207.
- Derntl, B., ., ., ., . . et al.** (2010). Multidimensional assessment of empathic abilities: neural correlates and gender differences. *Psychoneuroendocrinology*, 35, 67–82.
- Dodich, A., ., ., ., . et al.** (2014). Emotion recognition from facial expressions: a normative study of the Ekman 60-Faces test in the Italian population. *Neurological Sciences*, 35, 1015–21.
- Ekman, P. and Friesen, W. V.** (1975). *Pictures of Facial Affect*. Palo Alto, CA: Consulting Psychologists Press.
- Ekman, P., ., ., ., . et al.** (1987). Universals and cultural differences in the judgments of facial expressions of emotion. *Journal of Personality and Social Psychology*, 53, 712–7.
- Ekman, P., Sorenson, E. R. and Friesen, W. V.** (1969). Pan-cultural elements in facial displays of emotion. *Science*, 164, 86–8.
- Engelmann, J. B. and Pogossyan, M.** (2013). Emotion perception across cultures: the role of cognitive mechanisms. *Frontiers in Psychology*, 4, 118.
- Grainger, S. A., Henry, J. D., Phillips, L. H., Vanman, E. J. and Allen, R.** (2015). Age deficits in facial affect recognition: the influence of dynamic cues. *The Journals of Gerontology. Series B*, 72, 622–632.
- Halberstadt, J., Ruffman, T., Murray, J., Taumoepeau, M. and Ryan, M.** (2011). Emotion perception explains age-related differences in the perception of social gaffes. *Psychology and Aging*, 26, 133–6.
- Kalpourzos, G. et al.** (2009). Structural and metabolic correlates of episodic memory in relation to the depth of encoding in normal aging. *Journal of Cognitive Neuroscience*, 21, 372–89.
- Khawar, R., Malik, F., Maqsood, S., Yasmin, T. and Habib, S.** (2013). Age and gender differences in emotion recognition ability and intellectual functioning. *Journal of Behavioural Sciences*, 23, 53–71.
- Kumfor, F. and Piguet, O.** (2013). Emotion recognition in the dementias: brain correlates and patient implications. *Neurodegenerative Disease Management*, 3, 277–88.
- Lawrence, K., Campbell, R. and Skuse, D.** (2015). Age, gender, and puberty influence the development of facial emotion recognition. *Frontiers in Psychology*, 6, 761.
- Lindquist, K. A., Gendron, M., Barrett, L. F. and Dickerson, B. C.** (2014). Emotion perception, but not affect perception, is impaired with semantic memory loss. *Emotion*, 14, 375–87.
- Mill, A., Allik, J., Realo, A. and Valk, R.** 2009. Age-related differences in emotion recognition ability: a cross-sectional study. *Emotion*, 9, 619–30.
- Nitrini, R. et al.** (2004). Performance of illiterate and literate nondemented elderly subjects in two tests of long-term memory. *Journal of the International Neuropsychological Society*, 10, 634–8.
- Nitrini, R. et al.** (2009). Prevalence of dementia in Latin America: a collaborative study of population-based cohorts. *International Psychogeriatric*, 21, 622–30.
- Nitrini, R. and Caramelli, P.** (2003). Demências. In R. Nitrini and L. Bacheschi (eds.), *A Neurologia Que Todo Médico Deve Saber*. São Paulo: Atheneu.
- Opdebeeck, C., Martyr, A. and Clare, L.** (2016). Cognitive reserve and cognitive function in healthy older people: a meta-analysis. *Neuropsychology, Development, and Cognition. Section B, Aging, Neuropsychology and Cognition*, 23, 40–60.
- Roca, M. et al.** (2013). Intelligence and executive functions in frontotemporal dementia. *Neuropsychologia*, 51, 725–30.
- Ruffman, T., Henry, J. D., Livingstone, V. and Phillips, L. H.** (2008). A meta-analytic review of emotion recognition and aging: implications for neuropsychological models of aging. *Neuroscience and Biobehavioral Reviews*, 32, 863–81.
- Russell, J. A.** (1994). Is there universal recognition of emotion from facial expression? A review of the cross-cultural studies. *Psychological Bulletin*, 115, 102–41.
- Torralva, T., Roca, M., Gleichgerrcht, E., Bekinschtein, T. and Manes, F.** (2009). A neuropsychological battery to detect specific executive and social cognitive impairments in early frontotemporal dementia. *Brain*, 132, 1299–309.

Neuropsychological correlates of instrumental activities of daily living in neurocognitive disorders: a possible role for executive dysfunction and mood changes

Martina Amanzio,^{1,2} Sara Palermo,¹ Milena Zucca,³ Rosalba Rosato,^{1,4} Elisa Rubino,³ Daniela Leotta,⁵ Massimo Bartoli¹ and Innocenzo Rainero³

¹Department of Psychology, University of Turin, Via Verdi 10, 10123 Turin, Italy

²European Innovation Partnership on Active and Healthy Ageing, Bruxelles, Belgium

³Department of Neuroscience, University of Turin, Via Cherasco 15, 10126 Turin, Italy

⁴Unit of Cancer Epidemiology, Città della Salute e della Scienza Hospital and CPO Piemonte, Turin, Italy

⁵Martini Hospital, Neurology Division, Via Tofane 71, 10100 Turin, Italy

ABSTRACT

Since baseline executive dysfunction predicts worsening Instrumental Activities of Daily Living (i-ADL) over time and progression to Alzheimer's Disease (AD), we aimed to analyze the role of neuropsychological variables to outline which factors can contribute to functional impairment. Specific attention to executive functions (EFs) has been given.

A total of 144 subjects complaining of different cognitive deficits – ranging from “MCI likely due to AD” to “mild AD patients” – underwent an overall neuropsychological assessment. The *Behavioral Assessment of the Dysexecutive Syndrome* was used to analyze EFs. We conducted multiple linear regression analyses to study whether the level of independent living skills – assessed with the Lawton-scale – could be associated with cognitive and behavioral measurements.

We found a significant association between i-ADL and specific EFs measured by *Rule Shift Cards* ($p = 0.04$) and *Modified Six Elements* ($p = 0.02$). Moreover, considering i-ADL scores, we observed an involvement of mood changes and a reduced awareness of deficits in terms of Hamilton Depression Rating Scale ($p = 0.02$) and Awareness of Deficit Questionnaire – Dementia scale ($p < 0.0001$), respectively.

Our results suggest the importance of considering the association between a reduction in i-ADL and executive dysfunction in patients who have AD etiopathology, for which the ability to inhibit a response, self-monitoring, set-shifting and mood deflection play a key role. Besides, no straightforward associations between i-ADL scores and global cognition, memory, language comprehension, attention, and perspective taking abilities were found.

Key words: IADL, mild cognitive impairment, Alzheimer's disease, executive dysfunction

Introduction

The cognitive changes associated with neurodegenerative diseases, such as Alzheimer's Disease (AD), lead to a progressive decline in the patient's ability to perform activities of daily living (ADL). Recent evidence has suggested that AD is a continuum, with the clinical symptoms of a major neurocognitive disorder (DSM-5: American Psychiatric Association, 2013) becoming apparent a decade or more after the biomarker-associated

pathophysiological process begins in sporadic AD (Morris *et al.*, 2009; Rentz *et al.*, 2010; Sperling *et al.*, 2011; Knopman *et al.*, 2012; Ellis *et al.*, 2013; Villemagne *et al.*, 2013), and autosomal dominant AD (Bateman *et al.*, 2012). Research concerning biomarkers in the early stages of neurodegeneration has suggested that functional impairment occurs before cognitive impairment (Jack *et al.*, 2010).

ADL can be stratified according to difficulty and complexity in three levels of functioning (Reuben *et al.*, 1990). Basic ADL (b-ADL) are defined as the activities meeting the basic physiological and self-maintenance needs. Instrumental ADL (i-ADL) are essential, together with b-ADL, to maintain independent living. Advanced ADL (a-ADL) are more sophisticated activities, beyond those

Correspondence should be addressed to: Sara Palermo, Department of Psychology, University of Turin, Via Verdi 10, Turin 10124, Italy. E-mail: sara.palermo@unito.it. Received 22 Sep 2017; revision requested 29 Oct 2017; revised version received 26 Feb 2018; accepted 2 Mar 2018. First published online 23 May 2018.

necessary to live independently (De Vriendt *et al.*, 2012, 2015) such as, using (household) technology, driving, going on holidays, doing sports, practice hobbies, or arts (De Vriendt *et al.*, 2012).

i-ADL may be impaired in the early stage of AD (Marshall *et al.*, 2011a; 2014) and are more likely to be sensitive to the early effects of cognitive decline (Pérès *et al.*, 2008). Traditionally, general cognitive functioning (other than the presence of memory complaints) needed to be preserved – as well as the capability to perform daily life activities independently – for a person to be classified as having a Mild Cognitive Impairment (MCI) (Petersen *et al.*, 1999). In particular, when MCI was subsequently described as “*a concept in evolution*,” it was reported that very mild problems in i-ADL are generally consistent with MCI, while b-ADL should be preserved (Petersen *et al.*, 2014). Importantly, MCI associated with compromised i-ADL abilities has been found to predict progression to major neurocognitive disorders (Yoshita *et al.*, 2006; Ogama *et al.*, 2014, 2016; Jenkel *et al.*, 2015). Moreover, there is increasing evidence for early i-ADL decrements in individuals with amnesic MCI (aMCI) (Farias *et al.*, 2005; Bangen *et al.*, 2010; Luck *et al.*, 2011). This association is in line with the finding that aMCI represents an increased risk for major neurocognitive disorder, such as AD (Jungwirth *et al.*, 2012).

Impairment of daily life functions worsens with the clinical stage of AD, and increases caregivers' burden (Kamiya *et al.*, 2014). The following have been reported to be associated with i-ADL impairment: cognitive decline (Burton *et al.*, 2006; Cahn-Weiner *et al.*, 2007; Royall *et al.*, 2007; Tomaszewski *et al.*, 2009), depressive symptoms (Kondo *et al.*, 2008; Hybels *et al.*, 2009; Nyunt *et al.*, 2012; Song *et al.*, 2014), female sex (Sahin *et al.*, 2015), lower education (Sahin *et al.*, 2015), older age (Sahin *et al.*, 2015), physical dysfunction (Seidel *et al.*, 2011; Gobbens *et al.*, 2014; Albert *et al.*, 2015; Artaud *et al.*, 2015), and executive dysfunction (Burton *et al.*, 2006; Cahn-Weiner *et al.*, 2007). The evidence that executive dysfunction impacts i-ADL in subjects with AD was previously reported (Boyle *et al.*, 2003; Pereira *et al.*, 2008; Tomaszewski *et al.*, 2009). In this direction, a study by Marshall *et al.* (2011b) further demonstrated a significant relationship between executive dysfunction and i-ADL impairment in normal ageing, MCI, and mild AD. Executive dysfunction and i-ADL impairment have been shown to predict progression from aMCI to clinical AD (Tabert *et al.*, 2002). Moreover, they are thought to be associated with each other and prefrontal dysfunction (Tabert *et al.*, 2002). Interestingly, a reduced awareness of i-ADL deficits

brings patients with mild AD to overestimate their functional capacity. This aspect was previously associated with specific executive dysfunction – in terms of self-monitoring, set-shifting, response inhibition – and with the presence of mood changes (Amanzio *et al.*, 2011; 2013).

Since the association between functional impairment and executive dysfunction is important for diagnostic and prognostic purposes, we decided to further study this association taking into account specific aspects that, to the best of our knowledge, have not been analyzed in a single study: (1) A large group of participants had been carefully selected in order to represent subjects who have AD etiopathology as the cause of their impairments; (2) The cognitive deterioration had been studied, using an overall neuropsychological assessment, in order to analyze the contributions of different cognitive-behavioral sub-domains to functional dysfunctions; (3) Specific EFs have been analyzed in order to describe possible association with i-ADL disabilities. In particular, we conducted three multiple linear regression analyses in order to describe: (1) The role of global cognitive functioning and specific cognitive variables (selective attention, episodic memory, and language comprehension); (2) The role of specific executive functions (EFs), such as self-monitoring, set-shifting, and response inhibition; (3) The relationship with awareness of deficits, mood changes, and perspective taking in terms of Theory of Mind (ToM) of the first and second type (Premack and Woodruff, 1978).

Methods

Participants

All the outpatients were enrolled at the Neurology Division of the “Città della Scienza e della Salute” Hospital and the Martini Hospital, both in Turin (Italy).

Participants were included in the study if they had minor or major neurocognitive disorders (DSM-5, American Psychiatric Association, 2013), such as MCI likely due to AD and AD. Participants were excluded from the study if they had; (1) major depression or dysthymia, based on DSM-5 criteria (American Psychiatric Association, 2013); (2) a Mini-Mental State Examination (MMSE: Folstein *et al.*, 1975) score of <19 given that the neuropsychological measurement is not as reliable when problems of language comprehension occur; (3) were taking medications that could substantially impact cognitive functioning or antidepressants and/or anxiolytics. Cerebrospinal Fluid (CSF) diagnosis that did not provide *in-vivo* evidence of

Alzheimer's pathology was considered a further exclusion criterion.

The patients underwent extensive clinical and neuroradiological investigations, including structural magnetic resonance and Positron Emission Tomography with 2-deoxy-2-[fluorine-18] fluoro-D-glucose. Lumbar puncture with CSF measurement (phospho-Tau, total-Tau and $^{1-42}$ beta-amyloid) was performed on all patients (Innogenetics kits, Ghent, Belgium; see Table S1, available as supplementary material attached to the electronic version of this paper at www.journals.cambridge.org/jid_IPG). Diagnoses of MCI likely due to AD and of AD were based on the recommendations from the National Institute on Aging/Alzheimer's Association workgroups on diagnostic guidelines for AD (Albert *et al.*, 2011; McKhann *et al.*, 2011), taking into consideration the core research criteria (Dubois *et al.*, 2014).

Assessment of i-ADL

We verified the subjects' level of autonomy in daily living in terms of instrumental activities (Lawton and Brody, 1969) in the presence of a reliable informant. The scale was administered by interview to a knowledgeable family member or caregiver who provided answers.

The Lawton i-ADL scale is an appropriate instrument to assess independent living skills (Lawton and Brody, 1969). From a clinical perspective, it is emphasized that the i-ADL scale may provide an early warning of functional decline, or signal the need for further assessment (Graf, 2008). These skills are considered more complex than b-ADL. The instrument is most useful for identifying how a person is functioning at the present time and for identifying improvement or deterioration over time.

For all the above, the "Italian Society for Gerontology and Geriatrics" considers i-ADL scale as part of the Comprehensive Geriatric Assessment. Importantly, the Piedmont Welfare System considers i-ADL scale as having legal value during the health inspections at the Geriatric Assessment Units (D.G.R. n. 42-8390 10/3/2008).

The i-ADL scale (Lawton and Brody, 1969) evaluates functional autonomy in the performance of eight different functions; (1) using the telephone; (2) shopping; (3) preparing food; (4) housekeeping; (5) doing laundry; (6) using transportation; (7) handling medications; and (8) ability to handle finances. Each item was rated dichotomously (0 = less able, 1 = more able). Total scores range from 0 (low function, dependent) to 8 (high function, independent). The higher the score the lower the level of dependence. Each ability

measured by i-ADL scale relies on either cognitive or physical function, though all require some degree of both.

Neuropsychological assessment

The neuropsychological evaluation involved a wide assessment of global cognitive deterioration using: the MMSE and the Alzheimer's disease assessment scale – cognitive sub-scale (ADAS-cog: Rosen *et al.*, 1984). The disease severity was assessed with the Clinical Dementia Rating Scale (CDR: Hughes *et al.*, 1982), selective attention with Attentional Matrices (AM: Spinnler and Tognoni, 1987), episodic memory with the Recall of a Short Story test (Babcock: Spinnler and Tognoni, 1987), and language comprehension with the Token Test (De Renzi and Vignolo, 1962; TT: Spinnler and Tognoni, 1987). EFs were analyzed by means of the six subscales making up the behavioural assessment of the dysexecutive syndrome (BADS) neuropsychological battery (Wilson *et al.*, 1996). These can be summarized as follows: (1) The rule shift cards (RSC) subtest evaluates the ability to respond correctly to a rule and to shift from the use of one simple rule to another more complex one; (2) the action program (AP) assesses skills in solving a closed-ended sequential problem; (3) the key search (KS) subtest evaluates the ability to explore planning in the visual spatial domain and to solve an open-ended problem; (4) the temporal judgment (TJ) subtest ranks cognitive estimation ability; (5) the zoo map (ZM) subtest evaluates planning, sequential behavior, and the use of external feedback in problem solving; (6) the modified six elements (MSE) subtest evaluates ability to divide attention, task scheduling, performance monitoring, and prospective memory.

Importantly, as Lezak *et al.* (2004) pointed out, the BADS is the only test battery that is able to offer an extensive overview of EF analyses. Indeed, the BADS has been considered helpful in detecting executive dysfunction in a variety of diseases and in AD (Wilson *et al.*, 1996; Amanzio, *et al.*, 2008; Espinosa *et al.*, 2009; da Costa *et al.*, 2013). Moreover, performance on the BADS has already been found to be related to prefrontal activity (Rodrigues Gouveia *et al.*, 2007).

ToM of the first and second type, which refers to the "ability to mentalize," to understand the mental state of others and to predict behavior based on those states, was also assessed. As expressed by Premack and Woodruff (1978) "in saying that an individual has a ToM, we mean that the individual imputes mental states to himself and to others" (p. 515). In particular, ToM visual stories were used

to assess perspective-taking abilities (TOM 1 and TOM 2: Amanzio *et al.*, 2008). The subject has to solve problems involving: first-order attributions of false belief (of the type “A thinks X”) and second-order attributions of false belief (of the type “A thinks B thinks X”).

Specific neuropsychiatric scales for rating mood changes were also used to describe the patients’ behavioral profile: hypomania with the Mania Scale (MAS: Bech *et al.*, 1978); apathy and depression with the Hamilton Depression Rating Scale (HDR-S: Hamilton, 1960).

Unawareness of deficits at the time of testing was analyzed using the Awareness of Deficit Questionnaire – Dementia scale (AQ-D: Migliorelli *et al.*, 1995). The AQ-D is a scale of demonstrated reliability/validity for ranking the severity of unawareness of deficits in AD (Migliorelli *et al.*, 1995). Thirty questions divided into two sections (the cognitive and the behavioral) make up the questionnaire. The cognitive part assesses cognitive function and performance in ADL and i-ADL. The behavioral part assesses changes in interests and mood. All the questions were asked to the patients and to their caregivers blinded to the patients’ responses. Scores range from 0 (never) to 3 (always), with the minimum and maximum total scores obtainable ranging from 0 to 90 (cognitive section range = 0–66; behavioral section range = 0–24). The total AQ-D score is given by the difference between the caregiver’s and the patient’s forms. Higher scores on the AQ-D scale indicate greater unawareness of the disease and a reduced awareness of deficits, meaning that caregivers rated the patients as more impaired than did the patients themselves (Migliorelli *et al.*, 1995). Since this method is based on a subtractive index of perception (caregivers’ minus patients’ scores), the ruling out of any bias in the caregivers’ judgments is crucial (Amanzio *et al.*, 2011; 2013). For this reason, we have ensured that the caregivers (with a mean MMSE score of 27) had normal psychiatric and neurological evaluations and a negative history of neurological disorders.

Finally, b-ADL was assessed with the Katz *et al.* (1963) scale. The Katz Index of independence in ADLs is one of the most commonly used tools to assess basic ADLs (Milnac and Feng, 2016). It evaluates functional autonomy in the performance of six different functions: (1) bathing; (2) dressing; (3) toileting; (4) transferring; (5) continence; and (6) feeding. Each item was rated dichotomously (0 = dependent, 1 = independent). Total scores range from 0 (low function, dependent) to 6 (high function, independent). A score of 2 or less indicates severe functional impairment, 4 indicate moderate impairment, and 6 indicate

full function. Clinicians rate individuals as either fully independent (no supervision, direction, or personal assistance needed) or dependent (needing supervision, direction, personal assistance, or total care) across the six skills (Milnac and Feng, 2016). This measure was originally created to assess the physical functioning among those who were in rehabilitation (Milnac and Feng, 2016).

Procedures

Patients were evaluated by performing a neuropsychological assessment during their hospital admission lasting one week. The participants were assessed in three experimental sessions held one day apart and each lasting one-hour, with a view to preventing fatigue and lack of adherence to the tasks.

Statistical analysis

Statistical analyses were performed using SAS/STAT® 9.3 (Freund *et al.*, 1986; Schlotzhauer and Littell, 1987). Normality assumption distribution of outcome variables was evaluated using the Kolmogorov–Smirnov test. Since the distribution of i-ADL scores was not normal, we have dichotomized the variable using the median value (=6). We then divided the sample into above the median ($n = 62$) and up to the median ($n = 82$). The binary variable derived was afterwards used in multiple logistic regression analyses.

We conducted three logistic regression analyses adjusted for gender in order to study whether the level on i-ADL scale could be associated with cognitive and behavioral measurements. Importantly, we applied the “one in ten rule” according to which, logistic regression analyses could be used with a minimum of ten events per predictor variable (Harrell *et al.*, 1984; 1996).

Moreover, the selection of the three models was performed in line with the results obtained in the literature on i-ADL. In particular, i-ADL has been previously linked to general cognitive decline, specific cognitive functions, and neuropsychiatric domains (Marshall *et al.*, 2011b).

The final selected models considered i-ADL scale as the dependent variable and the following as independent variables (see Figure 1):

1. Model 1 – to address the role of global cognitive functioning and specific cognitive variables (global cognition, selective attention, episodic memory, and language comprehension): ADAS, AM, BABCOCK, TT.
2. Model 2 – to study the role of EFs with BADS sub-scales: BADS_RSC; BADS_AP; BADS_TJ; BADS_KS; BADS_ZOO; BADS_MSE.

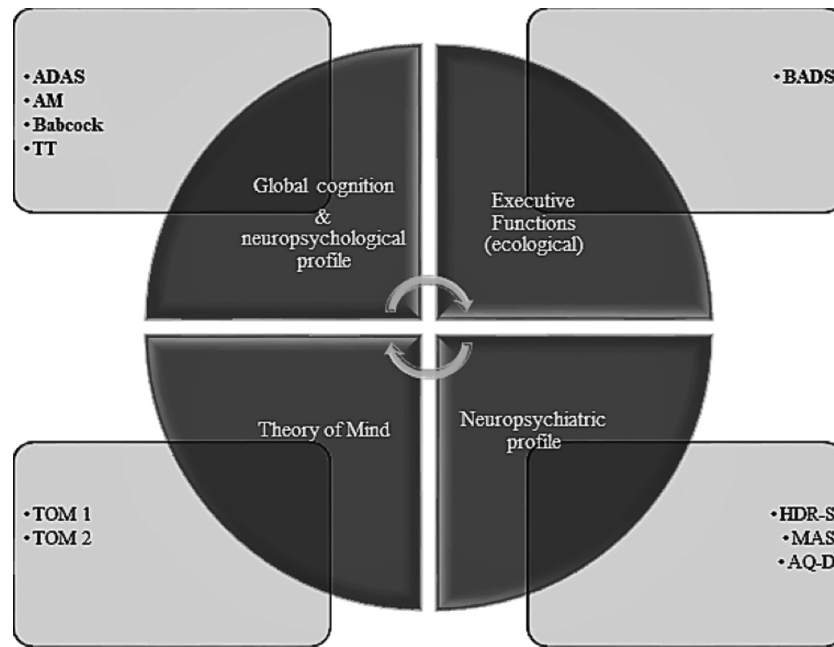


Figure 1. (Colour online) The graph represents the independent variables and the neuropsychological domain they belong to.

3. Model 3 – to investigate the relationship with awareness of deficits, mood changes, and TOM: AQ-D, HDR-S, MAS, TOM 1 and 2.

Results

Over a 26-month period, 200 patients – complaining of different cognitive deficits and presenting for the first time at the out-dep of our clinics – were evaluated for their possible participation in the study. Based on the inclusion criteria, 144 hospital admitted patients (M/F = 55/89; mean age \pm SD = 74.60 \pm 6.42 years) took part in the study. In particular, 32 subjects with MCI likely due to AD, according to the CSF analysis, were included in the study (see Table S1). For those patients with major neurocognitive disorders, the CSF diagnosis provided *in-vivo* evidence of Alzheimer's pathology for 112 patients. The demographic and clinical data related to the patient population have been summarized in Table 1. In anamnesis, only age-related disorders and problems (i.e. slight sensory deficits, hypertension, hypercholesterolemia, gastritis, weight gain or loss, and deflection of mood). All of them are drug-naïve patients. Indeed, they had not taken antidepressants and/or anxiolytics and/or anti-cholinesterase drugs before the neuropsychological evaluation. The neuropsychological assessment reflected the diagnoses made by the CSF, biomarkers and neurological exams. A total of 139 of the 144 patients obtained a CDR score of between 0 and 1,

Table 1. Demographic and clinical characteristics

| DEMOGRAPHIC CHARACTERISTICS | MEAN \pm SD |
|--|-------------------|
| Gender (male/female) | 55/89 |
| Age (years) | 74.60 \pm 6.42 |
| Schooling (years) | 7.83 \pm 3.57 |
| Early cognitive symptoms complaints (months) | 24.99 \pm 15.36 |
| CDR | 0.90 \pm 0.33 |
| <i>Functional assessment</i> | |
| b-ADL | 5.62 \pm 0.69 |
| i-ADL | 5.76 \pm 2.00 |
| i-ADL <6 N = 62 | |
| i-ADL >6 N = 82 | |

CDR = Clinical Dementia Rating Scale; SD = Standard Deviation.

b-ADL and i-ADL: higher scores indicate better performance.

indicating a low-level of cognitive impairment (see Table 2).

Considering the functional assessment, participants obtained a mean b-ADL score of 5.62 and a mean i-ADL score of 5.76, showing a low level of impairment on basic and instrumental ADL.

Association between i-ADL scale and neuropsychological variables

The results are presented as Odds Ratio (OR) and 95% confidence intervals (see Table 3). After adjusting the analysis for gender, i-ADL scores were influenced by BADS_RSC and BADS_MSE in model 2 and by AQ-D and HDR-S in model 3. Specifically, the worsening of performance at

Table 2. Neuropsychological and neuropsychiatric assessment synopsis. Wherever there is a normative value, the cut-off scores are given in the statistical normal direction. Cells in grey indicate the absence of a normative cut-off

| | MAXIMUM SCORE | MEAN \pm SD | CUT-OFF |
|--------------------------------------|---------------|-------------------|--------------|
| <i>Neuropsychological assessment</i> | | | |
| MMSE | 30 | 23.01 \pm 2.45 | ≥ 24 |
| ADAS | 100 | 20.74 \pm 7.08 | ≥ 82 |
| AM | 60 | 30.79 \pm 0.33 | ≥ 31 |
| Babcock | 16 | 3.49 \pm 3.48 | ≥ 4.75 |
| TT | 36 | 28.66 \pm 4.17 | ≥ 32.69 |
| TOM 1 | 4 | 3.34 \pm 0.98 | ≥ 3 |
| TOM 2 | 4 | 2.48 \pm 1.28 | ≥ 3 |
| BADS total score | 24 | 9.78 \pm 3.65 | ≥ 15 |
| Subtest RSC | 4 | 1.21 \pm 0.96 | |
| Subtest AP | 4 | 2.90 \pm 1.21 | |
| Subtest KS | 4 | 1.10 \pm 1.38 | |
| Subtest TJ | 4 | 1.63 \pm 1.01 | |
| Subtest ZM | 4 | 1.02 \pm 1.41 | |
| Subtest MSE | 4 | 1.92 \pm 0.80 | |
| <i>Neuropsychiatric Assessment</i> | | | |
| HDR-S | 67 | 7.35 \pm 4.91 | ≤ 7 |
| MAS | 44 | 2.78 \pm 2.80 | ≤ 15 |
| AQ-D total score | 90 | 16.67 \pm 16.33 | ≤ 14 |

MMSE = Mini-Mental State Examination; ADAS = Alzheimer's disease assessment scale; AM = Attentional Matrices; TT = Token Test; TOM = Theory of Mind; BADS = Behavioral Assessment of Dysexecutive Syndrome; RSC = Rule Shift Cards; AP = Action Program; KS = Key Search; TJ = Temporal Judgment; ZM = Zoo Map; MSE = Modified Six Elements; HDR-S = Hamilton Depression Rating Scale; MAS = Mania Scale; AQ-D = Awareness of Deficit Questionnaire – Dementia scale.

MMSE: lower scores indicate more severe cognitive impairment. ADAS: higher scores indicate more severe cognitive impairment. AM, Babcock, TT, TOM tasks and BADS: higher scores indicate better performance. AQ-D: higher scores indicate more severe unawareness. HDR-S and MAS scales: higher scores indicate more severe symptoms.

the BADS_RSC and BADS_MSE increases the probability that a participant has a dysfunction in i-ADL. Likewise, the chances of a subject being dysfunctional increases with the worsening of mood deflection and poor awareness. On the contrary, the level on the i-ADL scale was not influenced by global cognition, attention, memory, or language comprehension (in model 1).

Discussion

Our study is a first novel attempt to investigate possible association among i-ADL functioning, EFs and specific cognitive and behavioral variables, using an overall neuropsychological battery, in a selected patient population on the basis of CSF examination. Thus, newly diagnosed drug-naive MCI likely due to AD and AD patients provide an ideal population in which to study abnormalities in everyday functioning. Although we considered patients with different degrees of cognitive impairment, our sample was homogeneous in terms of etiopathogenesis, severity of symptoms (CDR = 0.90 ± 0.33 , attesting a mild level of

disease) and mood changes. Most importantly, our attempt to consider these kinds of patients in the same sample was justified by the regression analysis approach we used and by the international guidelines on aging that consider patients with cognitive impairment to lie on a continuum between MCI and mild AD (Petersen and Negash, 2008; Albert *et al.*, 2011; Dubois *et al.*, 2014).

Based on the results, we obtained, there appear to be no straightforward associations between i-ADL scores and specific aspects of neuropsychological functioning, such as global cognition, long-term verbal memory, language comprehension, and selective attention (in model 1). On the contrary, we observed a significant association between i-ADL functioning and two BADS sub-scales (in model 2), and between i-ADL and AQ-D and HDR-S (in model 3), respectively.

Our findings showed that i-ADL was associated with executive dysfunction. In particular, the ability to inhibit a response, self-monitoring, and set-shifting in terms of cognitive flexibility (measured through the MSE and the RSC) seem to be key skills for i-ADL, as demonstrated by the logistic regression analysis. As we previously

Table 3. Results for the logistic regression analysis applied in order to estimate the effect of neuropsychological and neuropsychiatric aspects on i-ADL. Outcomes were adjusted for the gender and are presented as Odds Ratio (OR) and 95% CI

| LAWTON-I-ADL | | PREDICTORS CONSIDERED ALL TOGETHER | | |
|--------------|----------|--|---------------|--------------------|
| | | EFFECTS | β | OR |
| MODEL 1 | ADAS | -0.055 | 0.947 | 0.084 |
| | AM | 0.001 | 1 | 0.984 |
| | BABCOCK | -0.005 | 0.995 | 0.935 |
| | TT | -0.029 | 0.971 | 0.593 |
| MODEL 2 | BADS_RSC | 0.472* | 1.603* | 0.044* |
| | BADS_AP | 0.363 | 1.437 | 0.052 |
| | BADS_KS | 0.034 | 1.035 | 0.837 |
| | BADS_TJ | -0.140 | 0.870 | 0.542 |
| | BADS_ZM | -0.300 | 0.742 | 0.135 |
| | BADS_MSE | 0.703* | 2.020 | 0.021* |
| MODEL 3 | AQ-D | -0.063 | 0.939* | <0.0001* |
| | TOM 1 | 0.473 | 1.605 | 0.093 |
| | TOM 2 | -0.225 | 0.799 | 0.308 |
| | HDR-S | -0.120* | 0.887* | 0.022* |
| | MAS | 0.069 | 1.072 | 0.428 |

*p < 0.05.

ADAS = Alzheimer's disease assessment scale; AM = Attentional Matrices; TT = Token Test; BADS = Behavioral Assessment of Dysexecutive Syndrome; BADS_RSC = subtest Rule Shift Cards; BADS_AP = subtest Action Program; BADS_KS = subtest Key Search; BADS_TJ = Temporal Judgment; BADS_ZM = Zoo Map; BADS_MSE = Modified Six Elements; AQ-D = Awareness of Deficit Questionnaire - Dementia scale; TOM = Theory of Mind; HDR-S = Hamilton Depression Rating Scale; MAS = Mania Scale.

reported (Amanzio *et al.*, 2013), being a modified version of Shallice and Burgess' Six Elements Test (Shallice and Burgess, 1991), the MSE was designed to assess the supervisory attentional system hypothesis. Specifically, MSE relies on the ability to inhibit a dominant response (i.e. perform the tasks in the given order), favoring the correct answer (i.e. alternating the execution of the tasks between all the proposed types). The MSE test also measures the ability to self-monitor performance and switch from task to task. In the same direction, the RSC subtest is a further measure of cognitive flexibility, involving the ability to move between different sets of responses. The RSC also measure abilities to shift and inhibit response and monitoring behavior (Cools *et al.*, 2000).

The three specific above-mentioned cognitive abilities – monitoring (updating), inhibition, and set-shifting – are defined in terms of basic EFs (Miyake *et al.*, 2000). These sub-components of executive control are considered mutually interacting (Miyake *et al.*, 2000). Not surprisingly,

all of them, if compromised, seem to be involved in functional disabilities. Interestingly, we previously demonstrated that executive dysfunction in terms of inhibition, self-monitoring, and set-shifting resulted associated with a reduction in the awareness of functional disabilities of mild AD patients (Amanzio *et al.*, 2013).

According to our results, planning or problem solving abilities – as higher-level subcomponent of EFs – did not seem to have any relationship with i-ADL. In particular, we found no positive results with BADS subtests, such as ZM, KS, and AP. Indeed, the ZM has been demonstrated to be useful in detecting planning impairment in AD. AD patients seem to have more problems developing logical strategies and executing complex predetermined plans (Piquard *et al.*, 2004; Allain *et al.*, 2007). KS is a more abstract task than the ZM, examining a person's ability to prepare an efficient plan of action in the context of a routine event. It is important to point out that these two tasks are considered to evaluate similar EFs (Wood and Liossi, 2007), related to the dorsolateral frontal lobe region (Millar *et al.*, 2006). In the same direction, AP assess the ability to develop an action-plan in order to solve a novel problem (Murakami *et al.*, 2015). Finally, we found no relationship between i-ADL and TJ measuring cognitive estimation ability (Murakami *et al.*, 2015).

Considering the neuropsychiatric profile and taking the IADL scores into account, we observed an involvement of mood changes, in terms of depression. This finding is consistent with the literature on minor and major neurocognitive disorders (Boyle *et al.*, 2003; Marshall *et al.*, 2011b). Moreover, patients with a reduced awareness of illness seem to have more difficulties in i-ADL. As we have previously demonstrated in AD patients, if the executive system does not function correctly, the comparator mechanism of self-monitoring does not detect mismatches between the current and previous performance states stored in the personal database and produces a reduced awareness for the instrumental domain (Amanzio *et al.*, 2013).

Finally, although AD patients may display TOM impairment primarily mediated by hippocampal degeneration (Synn *et al.*, 2018), we did not observed an association between functional impairment and mentalizing performance in our patients. Our results support the hypothesis that performances on social cognition tests are not a good indicator to differentiate patients with adequate i-ADL functioning from mild dysfunctioning patients.

Future prospective studies will be helpful in order to further characterize the role of

neuropsychological processes in the progression of i-ADL dysfunction.

Limitations section

The study here presented has been carefully designed and reached its aims; however, some critical aspects have to be outlined. The first aspect regards the tool used to assess the level of independent living skills, which could represent a possible confounding factor.

The evaluation of i-ADL may appear quite straight-forward. Despite this, procedures and tools can vary considerably. To date, a variety of examination tools measures the older adult's ADL and IADL performance, but there is no established gold standard for such assessment because few scales have been comprehensively evaluated (Capezuti *et al.*, 2017). Indeed, measures differ in their capability to establish level of dependence and the kind of assistance needed for each evaluated activity. Although Lawton i-ADL scale has low psychometrical properties (eventually affecting our results), it was developed to assess the more complex ADLs necessary for living in the community. This scale is part of the comprehensive geriatric assessment and is considered appropriate for use with older adults admitted to a hospital by the Italian legislation.

A second aspect concerns the results that have not to be considered generalizable for patients with different etiopathogenesis other than AD. However, our study was necessary to better define the associations between functional deficits and specific neuropsychological variables in a highly selected sample of patients.

Finally, we have focused our study in few predictors of IADL to assure a good power calculation. The selected predictors are in line with the international literature. Further studies would be necessary in order to analyze other factors.

Conclusion

Our results suggest the importance of considering EF dysfunctions in reduced i-ADL functionality in patients who have AD etiopathology as the cause of their impairments. The findings support the hypothesis that patients with different level of cognitive impairment, such as MCI likely due to AD and AD, exhibit i-ADL dysfunction in the context of overlapping EFs, reduced awareness of deficits and mood changes. A complete neuropsychological evaluation – based on specific assessment of the ability to inhibit a response, self-

monitoring, and set-shifting – might be able to identify those MCI patients, with reduced i-ADL functionality, at greater risk of developing a major neurocognitive disorder, such as AD. Finally, those patients with functional limitations in their daily living and reduced awareness may represent an important target population for tailoring specific interventions with important clinical implications, in terms of adherence to treatments and prognosis.

Conflict of interest

None.

Description of authors' roles

M. Amanzio designed the study, supervised the data collection, and wrote the paper. She took part in the review and critique processes as PI. She also organized the study and participated in the statistical analyses (execution and organization, review, and critique). S. Palermo supervised the data collection and the neuropsychological assessment, participated in the statistical analyses, participated in writing the paper, and created the infographics. R. Rosato was responsible for the statistical design of the study and for carrying out the statistical analysis and participated in writing the paper. E. Rubino performed the neurological assessment (execution) and took part in the organization of the study and in the diagnostic phase (organization and diagnosis). M. Zucca and M. Bartoli performed the neuropsychological assessment (execution). D. Leotta and I. Rainero supervised the neurological assessment, took part in the organization of the study, and participated in writing the paper (organization, review, and critique).

Acknowledgments

The study was approved by the Ethics Committee "A.O.U. Città della Salute e della Scienza di Torino - A.O. Ordine Mauriziano - A.S.L. Città di Torino" as part of the core research criteria followed by the Neurological Units. All subjects gave their informed written consent to participate in the study.

This study has therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Supplementary material

To view supplementary material for this article, please visit <https://doi.org/10.1017/S1041610218000455>

References

- Albert, M. S. et al.** (2011). The diagnosis of mild cognitive impairment due to Alzheimers disease: recommendations from the national institute on aging-Alzheimers association workgroups on diagnostic guidelines for Alzheimer's disease. *Alzheimer's & Dementia*, 7, 270–279. doi: 10.1016/j.jalz.2011.03.008.
- Albert, S. M., Bear-Lehman, J. and Anderson, S. J.** (2015). Declines in mobility and changes in performance in the instrumental activities of daily living among mildly disabled community-dwelling older adults. *Journal of Gerontology, Series A, Biological Science and Medical Science*, 70, 71–77. doi: 10.1093/gerona/glu088.
- Allain, P. et al.** (2007). A study of action planning in patients with Alzheimer's disease using the zoo map test. *Revue Neurologique*, 163, 222–230.
- Amanzio, M. et al.** (2011). Unawareness of deficits in Alzheimer's disease: role of the cingulate cortex. *Brain*, 134, 1061–1076. doi: 10.1093/brain/awr020.
- Amanzio, M., Geminiani, G., Leotta, D. and Cappa, S.** (2008). Metaphor comprehension in Alzheimer's disease: novelty matters. *Brain and Language*, 107, 1–10.
- Amanzio, M., Vase, L., Leotta, D., Miceli, R., Palermo, S. and Geminiani, G.** (2013). Impaired awareness of deficits in Alzheimer's disease: the role of everyday executive dysfunction. *Journal of the International Neuropsychological Society*, 19, 63–72. doi: 10.1017/S1355617712000896.
- American Psychiatric Association (APA)** (2013). *Diagnostic and Statistical Manual of Mental Disorders*, 5th edn. Washington, DC: American Psychiatric Association Publishing.
- Artaud, F., Singh-Manoux, A., Dugravot, A., Tzourio, C. and Elbaz, A.** (2015). Decline in fast gait speed as a predictor of disability in older adults. *Journal of American Geriatric Society*, 63, 1129–36. doi: 10.1111/jgs.13442.
- Bangen, K. J. et al.** (2010). Complex activities of daily living vary by mild cognitive impairment subtype. *Journal of the International Neuropsychological Society*, 16, 630–639. doi: 10.1017/S1355617710000330.
- Bateman, R. J. et al.** (2012). Clinical and biomarker changes in dominantly inherited Alzheimer's disease. *New England Journal of Medicine*, 367, 795–804. doi: 10.1056/NEJMoA1202753.
- Bech, P., Rafaelsen, O. J., Kramp, P. and Bolwig, T. G.** (1978). The mania rating scale: scale construction and inter-observer agreement. *Neuropharmacology*, 17, 430–431.
- Boyle, P. A., Malloy, P. F., Salloway, S., Cahn-Weiner, D. A., Cohen, R. and Cummings, J. L.** (2003). Executive dysfunction and apathy predict functional impairment in Alzheimer disease. *American Journal of Geriatric Psychiatry*, 11, 214–21.
- Burton, C. L., Strauss, E., Hultsch, D. F. and Hunter, M. A.** (2006). Cognitive functioning and everyday problem solving in older adults. *Clinical Neuropsychologist*, 20, 432–452. doi: 10.1080/13854040590967063.
- Cahn-Weiner, D. A. et al.** (2007). Cognitive and neuroimaging predictors of instrumental activities of daily living. *Journal of the International Neuropsychological Society*, 13, 747–757. doi: 10.1017/S1355617707070853.
- Capezuti, E. A., Malone, M. L., Khan, A. K. and Baumann, S. L.** (2017). *The Encyclopedia of Elder Care: The Comprehensive Resource on Geriatric Health and Social Care*, 4th edn. New York, NY: Springer Publishing Company.
- Cools, R., Brouwer, W. H., De Jong, R. and Slooff, C.** (2000). Flexibility, inhibition, and planning: frontal dysfunctioning in schizophrenia. *Brain and Cognition*, 108–112.
- da Costa Armentano, C. G., Porto, C. S., Nitrini, R. and Dozzi Brucki, S. M.** (2013). Ecological evaluation of executive functions in mild cognitive impairment and Alzheimer disease. *Alzheimer Disease & Associated Disorders*, 27, 95–101. doi: 10.1097/WAD.0b013e31826540b4.
- De Renzi, E. and Vignolo, L. A.** (1962). The token test: a sensitive test to detect receptive disturbances in aphasics. *Brain*, 85, 665–678.
- De Vriendt, P., Gorus, E., Cornelis, E., Velghe, A., Petrovic, M. and Mets, T.** (2012). The process of decline in advanced activities of daily living: a qualitative explorative study in mild cognitive impairment. *International Psychogeriatrics*, 24, 974–986. Doi: 10.1017/S1041610211002766.
- De Vriendt, P., Mets, T., Petrovic, M. and Gorus, E.** (2015). Discriminative power of the advanced activities of daily living (a-ADL) tool in the diagnosis of mild cognitive impairment in an older population. *International Psychogeriatrics*, 27, 1419–1427. Doi: 10.1017/S1041610215000563.
- Dubois, B. et al.** (2014). Advancing research diagnostic criteria for Alzheimer's disease: the IWG-2 criteria. *Lancet Neurology*, 13, 614–29. doi: 10.1016/S1474-4422(14)70090-0.
- Ellis, K. A. et al.** (2013). Decline in cognitive function over 18 months in healthy older adults with high amyloid. *Journal of Alzheimer's Disease*, 34, 861–871. doi: 10.3233/JAD-122170.
- Espinosa, A. et al.** (2009). Ecological assessment of executive functions in mild cognitive impairment and mild Alzheimer's disease. *Journal of the International Neuropsychological Society*, 15, 751–757. doi: 10.1017/S135561770999035X.
- Farias, S. T., Mungas, D. and Jagust, W.** (2005). Degree of discrepancy between self and other-reported everyday functioning by cognitive status: dementia, mild cognitive impairment, and healthy elders. *International Journal of Geriatric Psychiatry*, 20, 827–834.
- Folstein, M. F., Folstein, S. E. and McHugh, P. R.** (1975). "Mini-mental state". A practical method for grading the cognitive state of patients for the clinician. *Journal of Psychiatric Research*, 12, 189–198.
- Freund, R. J., Littell, R. C. and Spector, P. C.** (1986). *SAS System for Linear Models*. Cary, NC: SAS Institute Inc.
- Gobbens, R. J. and van Assen, M. A.** (2014). The prediction of ADL and IADL disability using six physical indicators of frailty: a longitudinal study in the Netherlands. *Current Gerontology and Geriatric Research*, 2014, 358137. doi: 10.1155/2014/358137.
- Graf, C.** (2008). The Lawton instrumental activities of daily living scale. *American Journal of Nursing*, 108, 52–62. doi: 10.1097/01.NAJ.0000314810.46029.74.
- Hamilton, M.** (1960). A rating scale for depression. *Journal of Neurology, Neurosurgery, and Psychiatry*, 23, 56–62.

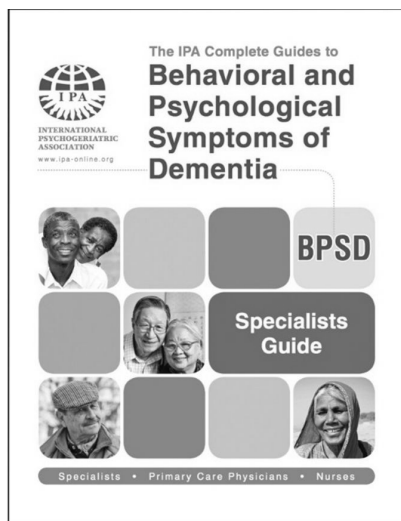
- Harrell, F. E., Lee, K. L., Califf, R. M., Pryor, D. B. and Rosati, R. A.** (1984). Regression modelling strategies for improved prognostic prediction. *Statistics in Medicine*, 3, 143–152.
- Harrell, F. E., Lee, K. L. and Mark, D. B.** (1996). Multivariable prognostic models: issues in developing models, evaluating assumptions and adequacy, and measuring and reducing errors. *Statistics in Medicine*, 15, 361–387.
- Hughes, C. P., Berg, L., Danziger, W. L., Coben, L. A. and Martin, R. L.** (1982). A new clinical scale for the staging of dementia. *British Journal of Psychiatry*, 140, 566–572.
- Hybels, C. F., Pieper, C. F. and Blazer, D. G.** (2009). The complex relationship between depressive symptoms and functional limitations in community-dwelling older adults: the impact of subthreshold depression. *Psychological Medicine*, 39, 1677–1688. doi: 10.1017/S0033291709005650.
- Jack, C. R. Jr. et al.** (2010). Hypothetical model of dynamic biomarkers of the Alzheimer's pathological cascade. *Lancet Neurology*, 9, 119–128. doi: 10.1016/S1474-4422(09)70299-6.
- Jekel, K. et al.** (2015). Mild cognitive impairment and deficits in instrumental activities of daily living: a systematic review. *Alzheimer's Research & Therapy*, 7, 17. doi: 10.1186/s13195-015-0099-0.
- Jungwirth, S., Zehetmayer, S., Hinterberger, M., Tragl, K. H. and Fischer, P.** (2012). The validity of amnesic MCI and non-amnesic MCI at age 75 in the prediction of Alzheimer's dementia and vascular dementia. *International Psychogeriatrics*, 24, 959–966. doi: 10.1017/S1041610211002870.
- Kamiya, M., Sakurai, T., Ogama, N., Maki, Y. and Toba, K.** (2014). Factors associated with increased caregivers' burden in several cognitive stages of Alzheimer's disease. *Geriatric & Gerontology International*, 14 (Suppl. 2), 45–55. doi: 10.1111/ggi.12260.
- Katz, S., Ford, A. B., Moskowitz, R. W., Jackson, B. A. and Jaffe, M. W.** (1963). Studies of illness in the aged. The index of Adl: a standardized measure of biological and psychosocial function. *JAMA*, 185, 914–916.
- Knopman, D. S. et al.** (2012). Short-term clinical outcomes for stages of NIA-AA preclinical Alzheimer disease. *Neurology*, 78, 1576–1582. doi: 10.1212/WNL.0b013e3182563bbe.
- Kondo, N., Kazama, M., Suzuki, K. and Yamagata, Z.** (2008). Impact of mental health on daily living activities of Japanese elderly. *Preventive Medicine*, 46, 457–462. doi: 10.1016/j.ypmed.2007.12.007.
- Lawton, M. P. and Brody, E. M.** (1969). Assessment of older people: self-maintaining and instrumental activities of daily living. *Gerontologist*, 9, 179–186.
- Lezak, M. D., Howieson, D. B. and Loring, D. W.** (2004). *Neuropsychological Assessment*, 4th edn. New York: Oxford University Press.
- Luck, T., Luppa, M., Angermeyer, M. C., Villringer, A., König, H. H. and Riedel-Heller, S. G.** (2011). Impact of impairment in instrumental activities of daily living and mild cognitive impairment on time to incident dementia: results of the Leipzig longitudinal study of the aged. *Psychological Medicine*, 41, 1087–1097. doi: 10.1017/S003329171000142X.
- Marshall, G. A. et al.** (2011a). Instrumental activities of daily living impairment is associated with increased amyloid burden. *Dementia and Geriatric Cognitive Disorders*, 31, 443–450. doi: 10.1159/000329543.
- Marshall, G. A. et al.** (2011b). Executive function and instrumental activities of daily living in mild cognitive impairment and Alzheimer's disease. *Alzheimer's & Dementia*, 7, 300–308. doi: 10.1016/j.jalz.2010.04.005.
- Marshall, G. A. et al.** (2014). Regional cortical thinning and cerebrospinal biomarkers predict worsening daily functioning across the Alzheimer disease spectrum. *Journal of Alzheimer's Disease*, 41, 719–728. doi: 10.3233/JAD-132768.
- McKhann, G. M. et al.** (2011). The diagnosis of dementia due to Alzheimer's disease: recommendations from the national institute on aging-Alzheimer's association workgroups on diagnostic guidelines for Alzheimer's disease. *Alzheimer's & Dementia*, 7, 263–269. doi: 10.1016/j.jalz.2011.03.005.
- Migliorelli, R. et al.** (1995). Anosognosia in Alzheimer's disease: a study of associated factors. *Journal of Neuropsychiatry and Clinical Neurosciences*, 7, 338–344.
- Millar, D., Griffiths, P., Zermansky, A. J. and Burn, D. J.** (2006). Characterizing behavioral and cognitive dysexecutive changes in progressive supranuclear palsy. *Movement Disorders*, 21, 199–207.
- Milnac, M. E. and Feng, M. C.** (2016). Assessment of activities of daily living, self-care, and independence. *Archives of Clinical Neuropsychology*, 31, 506–516. doi: 10.1093/arclin/acw049.
- Miyake, A., Friedman, N. P., Emerson, M. J., Witzki, A. H., Howerter, A. and Wagner, T. D.** (2000). The unity and diversity of executive functions and their contributions to complex “frontal lobe” tasks: a latent variable analysis. *Cognitive Psychology*, 41, 49–100. doi: 10.1006/cogp.1999.0734.
- Morris, J. C. et al.** (2009). Pittsburgh compound B imaging and prediction of progression from cognitive normality to symptomatic Alzheimer disease. *Archives Neurology*, 66, 1469–1475. doi: 10.1001/archneuro.2009.269.
- Murakami, et al.** (2015). Modified six elements test: earlier diagnosis of the correlation between motor and executive dysfunction in Parkinson's disease without dementia. *Neurology and Clinical Neuroscience*, 3, 209–214.
- Nyunt, M. S., Lim, M. L., Yap, K. B. and Ng, T. P.** (2012). Changes in depressive symptoms and functional disability among community-dwelling depressive older adults. *International Psychogeriatrics*, 24, 1633–1641. doi: 10.1017/S1041610212000890.
- Ogama, N., Sakurai, T., Shimizu, A. and Toba, K.** (2014). Regional white matter lesions predict falls in patients with amnesic mild cognitive impairment and Alzheimer's disease. *Journal of the American Medical Directors Association*, 15, 36–41. doi: 10.1016/j.jamda.2013.11.004.
- Ogama, N., Yoshida, M., Nakai, T., Niida, S., Toba, K. and Sakurai, T.** (2016). Frontal white matter hyperintensity predicts lower urinary tract dysfunction in older adults with amnesic mild cognitive impairment and Alzheimer's disease. *Geriatric & Gerontology International*, 16, 167–174. doi: 10.1111/ggi.12447.

- Pereira, F. S., Yassuda, M. S., Oliveira, A. M. and Forlenza, O. V.** (2008). Executive dysfunction correlates with impaired functional status in older adults with varying degrees of cognitive impairment. *International Psychogeriatrics*, 20, 1104–1115. doi: 10.1017/S1041610208007631.
- Pérès, K. et al.** (2008). Natural history of decline in instrumental activities of daily living performance over the 10 years preceding the clinical diagnosis of dementia: a prospective population-based study. *Journal of the American Geriatrics Society*, 56, 37–44.
- Petersen, R. C., Caracciolo, B., Brayne, C., Gauthier, S., Vesna Jelic, V. and Fratiglioni, L.** (2014). Mild cognitive impairment: a concept in evolution. *Journal of Internal Medicine*, 2014 Mar, 275, 214–228.
- Petersen, R. C. and Negash, S.** (2008). Mild cognitive impairment: an overview. *CNS spectrums*, 13, 45–53.
- Petersen, R. C., Smith, G. E., Waring, S. C., Ivnik, R. J., Tangalos, E. G. and Kokmen, E.** (1999). Mild cognitive impairment: clinical characterization and outcome. *Archives of Neurology*, 56, 303–308.
- Piquard, A., Derouesné, C., Lacomblez, L. and Siéroff, E.** (2004). Planning and activities of daily living in Alzheimer's disease and frontotemporal dementia. *Psychologie Neuropsychiatrie du Vieillessement*, 2, 147–156.
- Premack, D. and Woodruff, G.** (1978). Does the chimpanzee have a theory of mind?. *Behavioral and Brain Sciences*, 1, 515–526.
- Rentz, D. M. et al.** (2010). Cognition, reserve, and amyloid deposition in normal aging. *Annals of Neurology*, 67, 53–64. doi: 10.1002/ana.21904.
- Reuben, D. B., Laliberte, L., Hiris, J. and Mor, V.** (1990). A hierarchical exercise scale to measure function at the advanced activities of daily living (AADL) level. *Journal of the American Geriatrics Society*, 38, 855–861.
- Rodrigues Gouveia, P. A., Dozzi Brucki, S. M., Fleury Malheiros, S. M. and Bueno, O. F. A.** (2007). Disorders in planning and strategy application in frontal lobe lesion patients. *Brain and Cognition*, 63, 240–246.
- Rosen, W. G., Mohs, R. C. and Davis, K. L.** (1984). A new rating scale for Alzheimer's disease. *American Journal of Psychiatry*, 141, 1356–1364.
- Royall, D. R. et al.** (2007). The cognitive correlates of functional status: a review from the committee on research of the American neuropsychiatric association. *Journal of Neuropsychiatry and Clinical Neurosciences*, 19, 249–265. doi: 10.1176/jnp.2007.19.3.249.
- Sahin, A. et al.** (2015). Factors affecting daily instrumental activities of the elderly. *Turkish Journal of Medical Sciences*, 45, 1353–1359.
- Schlotzhauer, S. D. and Littell, R. C.** (1987). *SAS System for Elementary Statistical Analysis*. Cary, NC: SAS Institute Inc.
- Seidel, D., Brayne, C. and Jagger, C.** (2011). Limitations in physical functioning among older people as a predictor of subsequent disability in instrumental activities of daily living. *Age and Ageing*, 40, 463–469. doi: 10.1093/ageing/afr054.
- Shallice, T. I. M. and Burgess, P. W.** (1991). Deficits in strategy application following frontal lobe damage in man. *Brain*, 114, 727–741.
- Song, H. J., Meade, K., Akobundu, U. and Sahyoun, N. R.** (2014). Depression as a correlate of functional status of community-dwelling older adults: utilizing a short-version of 5-item geriatric depression scale as a screening tool. *Journal of Nutrition, Health & Aging*, 18, 765–770. doi: 10.1007/s12603-014-0452-1.
- Sperling, R. A. et al.** (2011). Toward defining the preclinical stages of Alzheimer's disease: recommendations from the national institute on aging-Alzheimer's association workgroups on diagnostic guidelines for Alzheimer's disease. *Alzheimer's & Dementia*, 7, 280–292. doi: 10.1016/j.jalz.2011.03.003.
- Spinnler, H. and Tognoni, G.** (1987). Standardizzazione italiana e taratura di test neuropsicologici. *Italian Journal of Neurological Sciences*. Milan: Masson Italia Periodici.
- Synn, A. et al.** (2018). Mental states in moving shapes: distinct cortical and subcortical contributions to theory of mind impairments in dementia. *Journal of Alzheimer's Disease*, 61, 521–535. doi: 10.3233/JAD-170809.
- Tabert, M. H. et al.** (2002). Functional deficits in patients with mild cognitive impairment: prediction of AD. *Neurology*, 58, 758–764.
- Tomaszewski, F. S. et al.** (2009). Longitudinal changes in memory and executive functioning are associated with longitudinal change in instrumental activities of daily living in older adults. *Clinical Neuropsychologist*, 23, 446–461. doi: 10.1080/13854040802360558.
- Villemagne, V. L. et al.** (2013). Amyloid β deposition, neurodegeneration, and cognitive decline in sporadic Alzheimer's disease: a prospective cohort study. *Lancet Neurology*, 12, 357–367. doi: 10.1016/S1474-4422(13)70044-9.
- Wilson, B. A., Alderman, N., Burgess, P. W., Emslie, H. and Evans, J. J.** (1996). *BADS: Behavioural Assessment of the Dysexecutive Syndrome*. Bury St. Edmonds, UK: Thames Valley Test Company.
- Wood, R. L. and Liossi, C.** (2007). The relationship between general intellectual ability and performance on ecologically valid executive tests in a severe brain injury sample. *Journal of the International Neuropsychological Society*, 13, 90–98.
- Yoshita, M. et al.** (2006). Extent and distribution of white matter hyperintensities in normal aging, MCI, and AD. *Neurology*, 67, 2192–2198. doi: 10.1212/01.wnl.0000249119.95747.1f.

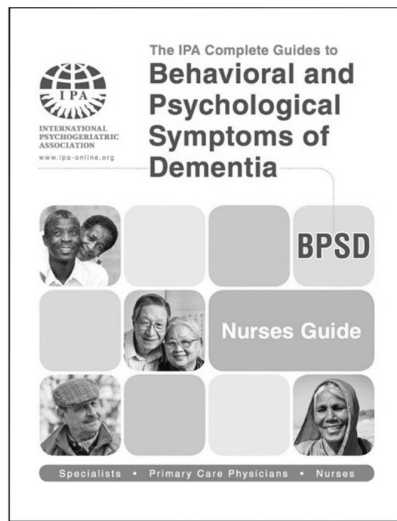


International Psychogeriatric Association
Better Mental Health for Older People

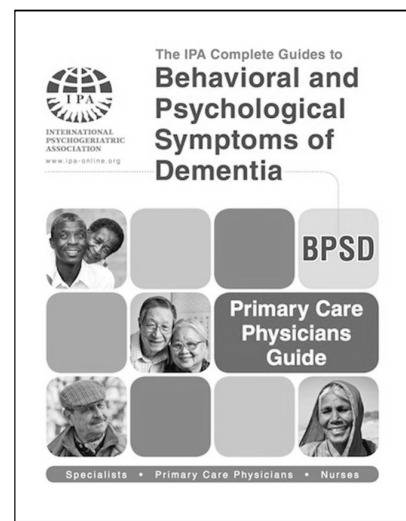
***The IPA Complete Guides to
Behavioral and Psychological Symptoms of Dementia (BPSD)***



Specialists Guide
Recently Revised!



Nurses Guide



**Primary Care
Physicians Guide**

The **Only** Resources You Need for **All** of the
Members of Your Team!

The IPA Complete Guides to Behavioral and Psychological
Symptoms of Dementia (BPSD) are available to all members of
the International Psychogeriatric Association (IPA).

To learn more about IPA and access the Guides, please visit:
www.ipa-online.org

The recency ratio as predictor of early MCI

Davide Bruno,¹ Rebecca L. Kosciak,² John L. Woodard,³ Nunzio Pomara^{4,5}
and Sterling C. Johnson^{6,2,7}

¹School of Natural Science and Psychology, Liverpool John Moores University, Liverpool, UK; Department of Psychology, Liverpool Hope University, Liverpool, UK

²Wisconsin Alzheimer's Institute, School of Medicine and Public Health, University of Wisconsin-Madison, Madison, WI, USA

³Department of Psychology, Wayne State University, Detroit, Michigan, USA

⁴Nathan Kline Institute for Psychiatric Research, Orangeburg, New York, USA; Department of Psychiatry, School of Medicine, New York University, New York City, New York, USA

⁵Department of Psychiatry, School of Medicine, New York University, New York City, New York, USA

⁶Geriatric Research Education and Clinical Center, Wm. S. Middleton Veterans Hospital, Madison, Wisconsin, USA

⁷Alzheimer's Disease Research Center, University of Wisconsin School of Medicine and Public Health, Madison, Wisconsin, USA

ABSTRACT

Objectives: Individuals with Alzheimer's disease (AD) present poor immediate primacy recall accompanied by intact or exaggerated recency, which then tends to decline after a delay. Bruno *et al.* (*Journal of Clinical and Experimental Neuropsychology*, Vol. 38, 2016, pp. 967–973) have shown that higher ratio scores between immediate and delayed recency (i.e. the recency ratio; Rr) are associated with cognitive decline in high-functioning older individuals. We tested whether Rr predicted conversion to early mild cognitive impairment (early MCI) from a cognitively healthy baseline.

Design: Data were analyzed longitudinally with binomial regression. Baseline scores were used to predict conversion to early MCI after approximately nine years. *Setting:* Data were collected at the Wisconsin Registry of Alzheimer's Prevention, in Madison, Wisconsin.

Participants: For the study, 427 individuals were included in the analysis; all participants were 50 years of age or older and cognitively intact at baseline, and were native English speakers.

Measurements: Memory data were collected using the Rey's Auditory Verbal Learning Test, and the early MCI diagnosis was obtained via consensus conference.

Results: Our results showed that higher Rr scores are correlated with greater risk of later early MCI diagnosis, and this association is independent of total recall performance.

Conclusions: Rr is an emerging cognitive marker of cognitive decline.

Key words: Alzheimer's disease, recency ratio, serial position, early MCI

Introduction

A common pattern in tests of human memory performance is the serial position curve, especially when memory is tested immediately after learning: performance is typically better for stimuli learned either at the beginning (primacy) or at the end (recency) of a study list, as compared to the middle (e.g. Murdock, 1962). The serial position curve assumes a particular shape for immediate-free recall tasks in individuals with Alzheimer's disease (AD), who present a reduction of the primacy effect,

while the recency effect is intact or exaggerated (e.g. Foldi *et al.*, 2003). However, when testing delayed performance, individuals with AD tend to show the most pronounced deficit at the recency position (Carlesimo *et al.*, 1995). Based on this discrepancy, Bruno *et al.* (2016) proposed that the ratio between immediate and delayed recency, i.e. the recency ratio (Rr), may measure cognitive decline. In particular, they proposed that higher ratios presented a pattern of enhanced immediate recency followed by loss of information after a time delay.

Although the exact neurocognitive mechanisms underlying the link between higher Rr scores and potential cognitive impairment are not entirely clear at this stage, Bruno *et al.* (2016) have proposed that individuals suffering consistent and

Correspondence should be addressed to: Davide Bruno, Ph.D., School of Natural Science and Psychology, Liverpool John Moores University Liverpool, UK. Phone: +44 (0)151 904 6320. Email: d.bruno@ljmu.ac.uk. Received 13 Oct 2017; revision requested 12 Jan 2018; revised version received 26 Jan 2018; accepted 5 Mar 2018. First published online 18 April 2018.

Table 1. Demographics

| | COGNITIVELY NORMAL | EARLY MCI | P VALUE |
|-------------------------------------|--------------------|-------------------|--------------------|
| N (%) | 367 (86%) | 60 (14%) | |
| Age at baseline | 56.6 (4.4; 50–68) | 57.3 (4.4; 50–65) | 0.234 |
| Time to follow up | 9.1 (1.0) | 9.2 (1.0) | 0.305 |
| Education | 5.0 (1.0) | 4.9 (1.1) | 0.600 |
| Females | 262 (71%) | 35 (58%) | 0.042 ^a |
| APOE ϵ 4/non- ϵ 4 | 132/235 | 22/38 | 0.917 ^a |
| AVLT total recall | 53.2 (6.8) | 46.5 (6.1) | <0.001 |
| Rr | 1.2 (0.6) | 1.3 (0.7) | 0.022 ^b |
| Delayed primacy | 0.8 (0.2) | 0.7 (0.3) | 0.002 ^b |

N = number of participants included in the analysis who either remained cognitively normal at follow up, or converted to early MCI; Age in years (mean, standard deviation, and range); Time to follow up in years (mean and standard deviation); Education level (median and range; from 1=8th grade or less to 6=post-graduate); Gender (number of females and percentage); APOE ϵ 4; AVLT total recall score at baseline (mean and standard deviation); Rr score at baseline (mean and standard deviation); and delayed primacy score at baseline (mean and standard deviation). Tests are *t*-tests unless specified. ^a χ^2 test was used; ^bMann–Whitney test was used.

severe loss of long-term memory and consolidation ability (e.g. individuals with dementia presumably due to AD) may rely more frequently on short-term memory processes, which, even if impaired, tend to be comparatively spared. Therefore, this long-to-short shift, possibly a compensatory mechanism, would naturally result in a pattern of performance consistent with higher Rr scores. To test this hypothesis, Bruno *et al.* examined whether Rr predicted changes in generalized cognitive ability (measured with the Mini-Mental State Examination, or MMSE; Folstein *et al.*, 1975) over two subsequent visits, from a cognitively healthy baseline. They observed that higher baseline Rr scores were correlated with more subsequent cognitive decline, and that decliners presented high immediate recency recall combined with a substantial drop in recency performance after a delay.

The present paper set out to confirm previous findings by testing whether Rr was associated with a subsequent diagnosis of early (preclinical) Mild Cognitive Impairment (early MCI; Kosciak *et al.*, 2016). The study was carried out over an average follow-up time of just over nine years (see Table 1), and all participants were cognitively intact at baseline. We anticipated that higher baseline Rr scores would predict greater risk of an early MCI diagnosis at the follow-up visit. To isolate the effects of Rr, we also controlled for total recall and delayed primacy effects (Bruno *et al.*, 2013).

Methods

Participants

Individuals were recruited as part of the Wisconsin Registry of Alzheimer Prevention (WRAP; Sager

et al., 2005). WRAP is a longitudinal study of participants who were middle-aged and free of clinical MCI or dementia at their baseline visit; participants complete follow-up visits, typically after two-to-four year intervals. Inclusion criteria for this study were that participants at baseline were not diagnosed by consensus conference as having any form of cognitive impairment (see the *Cognitive Status* section for details). Additionally, all participants were native English speakers, aged 50 years or over at baseline and had returned for follow up, receiving a consensus conference diagnosis classifying them as either cognitively normal or early MCI. Our final sample consisted of 427 participants, including 60 participants who converted to early MCI at follow up. The study was approved by the Health Sciences institutional review board of the University of Wisconsin–Madison, and the Faculty of Science Ethics committee at Liverpool Hope University.

Procedure

WRAP study procedures have been previously described in detail (e.g. Sager *et al.*, 2005). In brief, each visit included a neuropsychological test battery, and a series of self-report questionnaires on health history and lifestyle. In addition, blood was drawn for APOE genotyping (the procedure is described by Engelman *et al.* (2013)). The neuropsychological test battery included the Rey Auditory Verbal Learning Test (AVLT), where participants are read a list of 15 unrelated words before being asked to freely recall the items immediately (trial 1; i.e. the immediate recall trial). After the first learning trial, the same process is repeated four more times with the same words. Subsequently, a new list is read (interference), and participants once again are asked to recall the

original 15-word list. After a 20–25 min delay, participants are retested for their memory of the original word list (delayed recall trial). The same word list was used at all visits.

Cognitive Status

WRAP adopts a two-tiered consensus conference method to classify individuals in terms of their cognitive status. The first tier of review includes applying an algorithm that identifies cases where impairment may be present; and the second tier includes a team review of those flagged by the algorithm. Specifically, WRAP participant visits are reviewed at a consensus case conference if they meet one or more of the following criteria: (1) the participant is performing 1.5 SDs below the mean on factor scores or individual measures of memory, executive function, language, working memory, or attention (Koscik *et al.*, 2014; Clark *et al.*, 2016); (2) cognitive performance on one or more tests fell below values used in other studies as cut-points for clinical MCI diagnoses (e.g. WMS-R Logical Memory II, Wechsler, 1987: story A score <9: AD Neuroimaging Initiative, Petersen *et al.*, 2010); or (3) an abnormal informant report indicating subjective cognitive or functional decline. Consensus diagnoses of cognitively normal, early MCI, clinical MCI, dementia, and impaired-not-MCI are determined for each visit by a research team including physicians, clinical neuropsychologists, and clinical nurse practitioners based on review of cognitive, medical history, lifestyle, subjective cognitive complaints, and informant data (Koscik *et al.*, 2016). The status of early MCI was developed to identify individuals in the cohort who exhibit lower than expected objective performance in one or more cognitive domains relative to internal robust norms but do not report subjective cognitive complaints or clinical deficits. This experimental construct is thought to represent a phenotype of early cognitive decline expected to precede a clinical diagnosis of MCI. For the purposes of the present study, only individuals categorized as either cognitively normal or early MCI were included in the analysis. The exclusion of individuals with more severe classifications (e.g. dementia) was motivated by the desire to determine whether Rr was sensitive to the initial stages of disease progression and thus may be a potentially useful tool for early detection.

Serial Positions

Primacy and recency were defined as the first and last four items on the study list, respectively. Rr was calculated by dividing the recency scores in the immediate recall trial, Trial 1 of the AVLT,

by the corresponding scores in the delayed recall trial of the same test. An Rr score was calculated for each participant from the baseline visit data. A correction also was applied ((immediate recency score + 1)/(delayed recency score + 1)) to avoid missing data due to zero scores. Of note, this correction is different from the one used previously (Bruno *et al.*, 2016; 2017), since the original correction was found to generate paradoxical results.

Statistical Analysis

For the analysis, we performed a logistic regression with a binary outcome: the outcome was consensus diagnosis status at the follow-up visit, binarized to cognitively normal versus early MCI. We chose two time points for the analysis: baseline and a follow-up visit that took place at least seven years later. All participants were cognitively intact at baseline and either remained the same or converted to early MCI at follow up. Predictors were level of education (on a scale from 1, indicating 8th grade or less, to 6, indicating post-graduate studies); sex; APOE ϵ 4 status; time between baseline and follow up; Rr; delayed primacy (using primacy performance in the delayed trial); and total recall. To avoid issues of multi-collinearity, total recall was quantified here as the standardized residuals of total recall performance regressed over Rr; in turn, delayed primacy was similarly regressed over the total recall residuals and Rr together. Analyses were carried out in R version 3.2.3 (R Core Team, 2016), and SPSS 23 and 24.

Results

Table 1 reports means and standard deviations for the demographic variables, and memory scores. To confirm the suspicion of multi-collinearity, we ran bivariate correlations between Rr, total recall, and delayed primacy. Rr was significantly correlated with both total recall ($r = -0.249$, $p < 0.001$) and delayed primacy ($r = -0.256$, $p < 0.001$), which were in turn mutually correlated ($r = 0.465$, $p < 0.001$). These correlations were analogous using Spearman's ρ .

The analysis yielded two significant predictors: total recall (z value = -5.840 , $p < 0.001$), indicating that greater total recall was associated with lower risk of conversion to early MCI; and Rr (z value = 2.238 , $p = 0.025$), confirming the prediction that higher Rr scores are linked with greater risk of early MCI (other predictors, p 's > 0.24). Table 2 reports all regression results, including odds ratios. Of note, for every unit change in baseline Rr, the odds of an early MCI

Table 2. Output of the logistic regression analysis

| | UCE | SE | z VALUE | p VALUE | OR |
|-------------------------------|--------|-------|---------|---------|-----------------------|
| Intercept | -4.900 | 1.658 | -2.956 | 0.003 | 0.007 (<0.001, 0.186) |
| Time to follow up | 0.182 | 0.154 | 1.186 | 0.236 | 1.200 (0.885, 1.619) |
| Education | 0.080 | 0.152 | 0.524 | 0.600 | 1.083 (0.806, 1.467) |
| Sex | 0.121 | 0.335 | 0.362 | 0.717 | 1.129 (0.592, 2.208) |
| APOE ϵ 4 | -0.024 | 0.322 | -0.074 | 0.941 | 0.976 (0.513, 1.825) |
| Rr | 0.482 | 0.215 | 2.238 | 0.025 | 1.619 (1.046, 2.453) |
| AVLT total recall (residuals) | -1.080 | 0.185 | -5.840 | <0.001 | 0.340 (0.233, 0.482) |
| Delayed primacy (residuals) | 0.022 | 0.142 | 0.153 | 0.878 | 1.022 (0.777, 1.360) |

UCE = unstandardized coefficient estimate; SE = standard error; OR = odds ratios (confidence intervals: 2.5%, 97.5%).

classification later on increase (or decrease) by approximately 62%.

For the purposes of identifying potentially useful Rr cut-off points for clinical screening purposes, we note that whereas only 20% of early MCI converters (12/60) had an Rr score above 1.65, 85% of non-converters had an Rr score below 1.65 (308/367) – for a positive predictive value of 17%, and a negative predictive value of 87%. In contrast, 82% of converters had an Rr score at 1 or greater (49/60), but only 34% of non-converters had an Rr score below 1 (123/367) – for a positive predictive value of 17%, and a negative predictive value of 92%.

Discussion

In this paper, we aimed to expand on a previous report by Bruno *et al.* (2016) by examining serial position ratios in conjunction with diagnosis of early MCI. With binomial regression analysis, we found that the probability of receiving a diagnosis of early MCI was higher when the Rr score also was higher. Rr is based on recency performance, which focuses on memory for only the most recently presented information. At the immediate trial, this information has been presented only seconds prior, whereas in the delayed trial, 15–20 min have elapsed. Therefore, a high Rr score, and generally a score above 1, indicates that the person remembers comparatively more items immediately after learning than they do after a delay. Higher scores are suggestive of more forgetting over time as compared to lower scores, but such scores are considered in the context of a stronger performance in the immediate task. Bruno *et al.* (2016) have argued that shifting the emphasis from long-term retention (delayed performance) to short-term memory ability (immediate performance), particularly when evaluating recency performance, may be indicative of a compensatory mechanism whereby increased

long-term forgetting, presumably due to a loss of consolidation ability, leads to enhanced short-term memory processing. Therefore, paradoxically, improved performance at immediate recall is likely to fit into a negative cognitive profile signaling impending risk of cognitive decline, as our results suggest. Some evidence supporting this account comes from a recent report by Bruno *et al.* (2017) showing that Rr, but no other measure of memory in the study, was associated with levels of glutamate in the cerebrospinal fluid of a group of individuals with late-life major depression. Glutamate, the principal excitatory neurotransmitter in the brain, is implicated in long-term potentiation and the formation of long-lasting, consolidated memories. Specifically, consistent with the notion that a compensatory mechanism may emerge when cognitive ability deteriorates, Bruno *et al.* (2017) showed that whereas delayed recency increased when higher levels of glutamate were detected, the opposite was true for immediate recency; in other words, individuals whose glutamate levels were found to be higher appeared to rely less on short-term memory processing and more on long-term processing. More evidence is needed to elucidate this point, including whether this mechanism may be automatic or deliberate.

Despite the fact that baseline Rr was predictive or early MCI risk at follow up, we noted that the baseline total AVLT score yielded a stronger effect. This finding is not surprising because, although not exclusively, evaluation of broad AVLT categories is employed as part of the diagnostic process (see cognitive status), whereas Rr, albeit derived from the same test, is not. Nevertheless, Rr was shown to provide a predicting value for early MCI conversion above and beyond that of total AVLT. More importantly, in our view, is the fact that Rr may be narrowing in on specific mnemonic processes that are affected in AD, as opposed to the AVLT total recall score, which, as is a less specific index of memory performance, would likely include a number of different mnemonic processes that could be affected

by a host of different pathologies. Moving forward, it would be helpful to identify areas in which Rr may provide unique contributions to early detection and diagnosis of neurodegenerative disorders, including differential diagnosis of dementia types.

Delayed primacy performance was not predictive of early MCI conversion in this study. This finding may appear to contradict previous reports suggesting that delayed primacy was sensitive to subsequent cognitive decline (Bruno *et al.*, 2013), much like Rr. However, as noted, we employed in the analysis the residuals of delayed primacy regressed on Rr and the residuals of total recall. Therefore, in this instance, delayed primacy was only used as a control variable, and may not have been fairly represented. To confirm this point, we re-ran the analysis by replacing Rr with delayed primacy, and replacing the standardized residuals of total recall calculated from regressing total recall on Rr by the standardized residuals of total recall calculated from regressing total recall on delayed primacy, and further by adding standardized residuals of Rr calculated from delayed primacy and the residuals of total recall. The results show that delayed primacy is also predictive of early MCI conversion (unstandardized *coefficient* = -0.546 , *SE* = 0.163 , *z value* = -3.354 , $p < 0.001$), when controlling for total recall and Rr with an odds ratio of 0.579 (2.5–97.5% CIs = 0.420 – 0.797). Further research is needed to elucidate the different predictive values and underlying mechanisms of both primacy and recency recall performance.

All in all, our results suggest that serial position markers offer predictive value for the early identification of early MCI, independently from traditional neuropsychological measures of memory ability, such as total recall, and can therefore add to the array of cognitive markers for studies of neurodegenerative disorders. In this respect, we believe that researchers working on developing databases of AD biomarkers should consider including serial position values to their variables.

Conflicts of interest

None.

Description of author's roles

DB provided the concept, analyzed the data, and wrote the paper; RLK and JLW provided feedback on the paper, and on the statistical analysis in particular; NP helped developing the concept; and SCJ provided input on the clinical procedures, in addition to overseeing WRAP.

Acknowledgments

We wish to thank the WRAP study team for their data collection efforts and the WRAP participants for their dedication to the study. WRAP is supported by NIA grant R01AG27161 (SCJ). WRAP is also supported by the Clinical and Translational Science Award (CTSA) program, through the NIH National Center for Advancing Translational Sciences (NCATS), grant UL1TR000427. The content is solely the responsibility of the authors and does not necessarily represent the official views of the NIH.

References

- Bruno, D. et al.** (2015). A study on the specificity of the association between hippocampal volume and delayed primacy performance in cognitively intact elderly individuals. *Neuropsychologia*, *69*, 1–8.
- Bruno, D. et al.** (2017). The recency ratio is associated with reduced CSF glutamate in late-life depression. *Neurobiology of Learning and Memory*, *141*, 14–18.
- Bruno, D., Reichert, C. and Pomara, N.** (2016). The recency ratio as an index of cognitive performance and decline in elderly individuals. *Journal of Clinical and Experimental Neuropsychology*, *38*, 967–973.
- Bruno, D., Reiss, P. T., Petkova, E., Sidtis, J. J. and Pomara, N.** (2013). Decreased recall of primacy words predicts cognitive decline. *Archives of Clinical Neuropsychology*, *28*, 95–103.
- Carlesimo, G. A., Sabbadini, M., Fadda, L. and Caltagirone, C.** (1995). Different components in word-list forgetting of pure amnesics, degenerative demented and healthy subjects. *Cortex*, *31*, 735–745.
- Clark, L. R. et al.** (2016). Mild cognitive impairment in late middle age in the Wisconsin Registry for Alzheimer's Prevention (WRAP) study: prevalence and characteristics using robust and standard neuropsychological normative data. *Archives of Clinical Neuropsychology*, *31*, 675–688.
- Dowling, N. M., Hermann, B., La Rue, A. and Sager, M. A.** (2010). Latent structure and factorial invariance of a neuropsychological test battery for the study of preclinical Alzheimer's disease. *Neuropsychology*, *24*, 742.
- Engelman, C. D. et al.** (2013). Interaction between two cholesterol metabolism genes influences memory: findings from the Wisconsin registry for Alzheimer's Prevention. *Journal of Alzheimer's Disease*, *36*, 749–757.
- Foldi, N. S., Brickman, A. M., Schaefer, L. A. and Knutelska, M. E.** (2003). Distinct serial position profiles and neuropsychological measures differentiate late life depression from normal aging and Alzheimer's disease. *Psychiatry Research*, *120*, 71–84.
- Folstein, M. F., Folstein, S. E. and McHugh, P. R.** (1975). "Mini-mental state". A practical method for grading the cognitive state of patients for the clinician. *Journal of Psychiatric Research*, *12*, 189–198.

- Koscik, R. L. *et al.*** (2014). Emergence of mild cognitive impairment in late middle-aged adults in the Wisconsin registry for Alzheimer's prevention. *Dementia and Geriatric Cognitive Disorders*, 38, 16–30.
- Koscik, R. L. *et al.*** (2016). Intraindividual cognitive variability in middle age predicts cognitive impairment 8–10 years later: results from the Wisconsin registry for Alzheimer's prevention. *Journal of the International Neuropsychological Society*, 22, 1016–1025.
- Murdock, B. B.** (1962). The serial position effect of free recall. *Journal of Experimental Psychology*, 64, 482–488.
- Petersen, R. C. *et al.*** (2010). Alzheimer's disease Neuroimaging Initiative (ADNI) clinical characterization. *Neurology*, 74(3), 201–209.
- R Core Team** (2016). *R: A language and environment for statistical computing*. Vienna, Austria: R Foundation for Statistical Computing. Available at: <https://www.R-project.org/>
- Sager, M. A., Hermann, B. and La Rue, A.** (2005). Middle-aged children of persons with Alzheimer's disease: APOE genotypes and cognitive function in the Wisconsin registry for Alzheimer's prevention. *Journal of Geriatric Psychiatry and Neurology*, 18, 245–249.

Impact of brief education on healthy seniors' attitudes and healthcare choices about Alzheimer's disease and associated symptoms

Robyn E. Waxman,^{1,2,3} Barbara J. Russell,^{1,2,4} Oscar C. T. Lu² and Benoit H. Mulsant^{1,2}

¹Department of Psychiatry, University of Toronto, Toronto, Ontario, Canada

²Centre for Addiction and Mental Health, Toronto, Ontario, Canada

³Ontario Shores Centre for Mental Health Sciences, Whitby, Ontario, Canada

⁴University Health Network, Toronto, Ontario, Canada

ABSTRACT

Objective: The primary objective of this study was to determine whether a brief education session about Alzheimer's disease (AD) stages and associated behavioral and psychological symptoms of dementia (BPSD) changes healthy seniors' treatment choices. A secondary objective was to determine whether pharmacotherapy to reduce BPSD would be preferred over other potentially more restrictive interventions.

Methods: Participants ($n = 32$; 8 men; aged > 64 years; no self-reported dementia diagnosis) were assigned to one of ten group sessions during which they received information about AD and BPSD. Our a-priori hypotheses were: (1) education about AD stages significantly changes care preferences in moderate and severe stages, i.e. less active treatment options (no CPR/hospitalization) are chosen as the disease progresses; and (2) most participants prefer pharmacotherapy over restraints and seclusion to manage BPSD. The main outcome measure was a change in the interventions chosen including CPR and hospitalization. Participants completed three questionnaires and two decisional grids before and after the information session. Qualitative data were derived from discussions during the session.

Results: Participants expressed a wide range of attitudes about AD, BPSD, and their management. Those who are born in Canada, had a proxy, and a university education, each have around half of the odds of receiving treatment compared to those in the complementary group. (OR 0.47, 0.40, 0.43) Finally, not knowing someone with AD increases the odds of wanting a treatment by around six times (OR 6.4). Pharmacological measures were preferred over restraints.

Conclusions: Education about dementia and advance directives should consider the person's educational background and experience with dementia. Discussing BPSD may impact a person's advance directives and preferences.

Key words: Alzheimer's disease, behavioral and psychological symptoms of dementia, advance directives

Introduction

An estimated 5.2 million Americans have Alzheimer's disease (AD) and by 2050 the total estimated prevalence is expected to be 13.8 million. (Thies and Bleil, 2013) Clinicians have only recently acknowledged that AD qualifies as a terminal illness in addition to a progressively debilitating condition. (Morrison and Siu, 2000)

However, most of the general public does not see AD as a cause of death. Public knowledge of AD symptoms is often limited to memory loss and wandering (Gjerdingen *et al.*, 1999; Arai *et al.*, 2008). Behavioral and psychological symptoms of dementia (BPSD) are less well-known and can be more distressing and challenging. BPSD include disinhibition (e.g. undressing in public), physical or verbal aggression, high anxiety, and psychosis (e.g. delusional beliefs) (Margallo-Lana *et al.*, 2001; Lyketsos *et al.*, 2002). These symptoms present in 50% to 90% of people diagnosed with AD (Hersch and Falzgraf, 2007). The symptoms are associated with increased functional disability, earlier institutionalization, increased

Correspondence should be addressed to: Robyn E. Waxman, Ontario Shores Centre for Mental Health Sciences, 700 Gordon Street, Whitby, Ontario L1N 5S9, Canada. Phone: +(416) 450-1330; Fax: +(905) 430-4465. Email: waxmanro@ontarioshores.ca. Received 20 Oct 2017; revision requested 22 Nov 2017; revised version received 4 Mar 2018; accepted 5 Mar 2018. First published online 3 May 2018.

psychotropic medication use, decreased quality of life for caregivers and patients alike, and higher mortality rates (Madhusoodanan, 2001; Hersch and Falzgraf, 2007). Some non-pharmacological interventions can reduce BPSD, though admittedly they are not effective for every person (Leone *et al.*, 2009; Kolanowski *et al.*, 2010). Moreover, pharmacological interventions managing BPSD may be associated with serious adverse effects, including increased risk of mortality, falls, and over-sedation. Yet, alternatives to pharmacotherapy for significant aggression, such as physical and environmental restraints, can be traumatizing for the person to experience and family members to witness.

Busy clinicians often address BPSD only when symptoms emerge and become distressing rather than during the initial diagnosis and treatment phase. However, patient involvement and autonomy may be reduced further when they become incapable of making treatment decisions. Empirical studies have shown that in the absence of detailed advance directives or discussions, substitute decision makers – usually family members – often inaccurately choose treatment options preferred by the patient, despite their good intentions (Shalowitz *et al.*, 2006; Boustani *et al.*, 2008; Fried *et al.*, 2011). Physicians are frequently inaccurate as well (Uhlmann *et al.*, 1988; Hamann *et al.*, 2010). Advance directives are quasi-legal documents wherein people can state their preferences about who should serve as their substitute decision maker and which treatments and care should be continued, initiated, and discontinued. A successful advance care planning program was introduced in the United States in 2015 under medicare, which has resulted in 575,000 (double the expected number) Medicare patients discussing end-of-life issues with their families and physicians (Aleccia, 2017). Despite this number, only about 10% of people who develop dementia have written directives (Yung *et al.*, 2010), which often address only familiar life-sustaining measures such as artificial ventilation and nutrition, dialysis, or cardiopulmonary resuscitation (CPR) in case of organ failure, major neurological events, or cardiopulmonary arrests (Rurup *et al.*, 2005) without written directives for the management of serious symptoms like BPSD.

Studies have shown that education about the cognitive and functional decline associated with the progression of dementia can alter people's preferences for or against life-saving medical interventions (Deep *et al.*, 2010). However, the authors were unable to find a published study examining the impact of education addressing the impact of BPSD on seniors' preferences for medical interventions. Thus, we designed a study to examine the impact

of brief education about AD stages and BPSD on the treatment preferences of older adults free of dementia. We also explored their preferences for different management approaches of BPSD and their guiding reasons. We hypothesized that: (1) brief education about AD stages significantly changes treatment/care choices for moderate and severe stages, as evidenced by choosing less active treatment options (i.e. no CPR/hospitalization) as the disease progresses; and (2) most participants would prefer pharmacological options to manage BPSD, despite their risks.

Methods

Participants

This study was conducted in Toronto, a large, multicultural Canadian city, from June 2013 to July 2015.

The target population was adults older than 64 years of age. Participants were eligible if they reported being literate in English and not having a diagnosis of dementia. Posters about the study were displayed at one academic psychiatric hospital and three community senior centers. The research team also contacted people in the hospital's research registry. Interested individuals were informed of the study procedures, risks, and safeguards and mailed a consent form. They and those who enrolled were scheduled in groups of 2–6 people; in total 10 group sessions were held. If participants were family members or close friends, they were assigned to different groups to avoid possible undue influence of their responses and to maintain confidentiality. Accordingly, data from two participants were excluded from analyses when it was learned that they were married yet participated in the same small group.

Study design

A mixed methods approach was used. Demographic information included age, gender, education level, ethnicity, immigrant versus Canada-born background, marital and family status, religious/spiritual affiliation, personal knowledge or experience with family or friends having AD, and formal designation of a proxy decision maker.

In addition to the demographics questionnaire, participants completed two questionnaires prior to a brief education session: an abbreviated version of the AD Knowledge Scale (10 of the 30 original questions focused on diagnosis, prognosis, and management of AD; see Appendix 1, available as supplementary material attached to the electronic version of this paper at

| | | | |
|-------------------------------------|---|---|---|
| Kinds of treatment | Cardiopulmonary resuscitation & ventilation | Being hospitalized for tests and/or treatment | Relief of pain and symptoms at home |
| Your state of health... | | | |
| As it is today | | | |
| If you have mild dementia | | | |
| If you have moderate dementia | | | |
| If you have severe dementia | | | |

Figure 1. Decision grid 1. What would you want and not want?

| | | | | |
|--|--|---|--|---|
| Kinds of interventions | Cardio- pulmonary resuscitation & ventilation | Being hospitalized for tests and/or treatment | Relief of pain and symptoms at home | Rank order the following in terms of your preferences 1 = most preferable 2 = next most preferable 3 = least preferable |
| You have... | | | | |
| the BPSD symptom you think is the most undesirable | | | | ___ medications ___ physical restraints ___ seclusion |

Figure 2. Decision grid 2. What would you want and not want?

www.journals.cambridge.org/jid_IPG) (Carpenter *et al.*, 2009) and the Health Belief Model Questionnaire (HBMQ) adapted for AD (seven questions about attitudes and feelings about AD; see Appendix 2) (Janz and Becker, 1984; National Cancer Institute, 2011). These two questionnaires were completed again immediately after the brief education session and then one month later (via mail).

Participants also completed two decision grids. Decision grid #1 (see Figure 1) was completed before and after the education session. It focused on preferences for three common treatment plans in four health conditions: current state of health, mild AD, moderate AD, and severe AD. The three treatment plans were CPR and ventilation, hospitalization for testing and in-patient treatment, and at-home comfort measures for medical issues that may arise during the course of the AD.

Decision grid #2 (see Figure 2) was completed after the education session. First, it focused on

issues related to BPSD symptoms, specifically disinhibition, aggression, apathy, psychosis, and mood changes. Participants were asked to identify which of these symptoms was personally the most worrisome to live with and whether this symptom would change their preferences for and against the three aforementioned treatment plans. Finally, participants were asked to prioritize interventions that can be used to manage BPSD: physical restraints, environmental restraint (i.e. seclusion), and, pharmacotherapy (Narang *et al.*, 2010; Kim *et al.*, 2011; Stevens *et al.*, 2014).

One of the authors (REW) led most of the education session. The session lasted about 20 min, and consisted of a description of AD's progression from mild to moderate to severe stages using a case, highlighting changes in one's trajectory from semi-independence to complete dependence for Activities of Daily Living, the expected impact of AD on longevity and physical and cognitive abilities, and finally, available health treatments

Table 1. Participants' characteristics

| | | N | % |
|--|--------------------|------------|------|
| Gender | Female | 24 | 75.0 |
| | Male | 8 | 25.0 |
| Age | 65–71 years | 9 | 28.1 |
| | 72–79 years | 14 | 43.8 |
| | Over 79 years | 9 | 28.1 |
| | Mean (SD), years | 74.8 (5.6) | |
| Holds a university degree | Yes | 17 | 53.1 |
| | No | 15 | 46.9 |
| Marital status | Single | 8 | 25.0 |
| | Widowed | 7 | 21.9 |
| | Divorced/Separated | 7 | 21.9 |
| | Married/Common law | 10 | 31.3 |
| Canadian born | Yes | 15 | 46.9 |
| | No | 17 | 53.1 |
| Has a proxy for healthcare decisions | Yes | 21 | 65.6 |
| | No | 9 | 28.1 |
| | No response | 2 | 6.3 |
| Is close or has been close to a person with dementia | Yes | 29 | 90.6 |
| | No | 3 | 9.4 |

for conditions that may arise in the course of illness, their effectiveness, and potential risks. The session also included a review of five major BPSDs: psychosis, aggression, apathy, disinhibition, and mood changes. The education session was followed by a facilitated discussion of the reasons for expressed preferences, choices of treatment/care options, and any changes following the education session.

The study was reviewed and approved by three hospitals' Research Ethics Boards. All participants completed an approved written consent form prior to engaging in any of the study's procedures.

Data analysis

Quantitative data were derived from the demographics questionnaire, the shortened AD knowledge questionnaire, the adapted HBMQ, and responses on grid #1 and grid #2. An Omnibus test was used to assess changes in knowledge about AD before and after the education sessions and ordinal logistic regression models were used to assess for the effect of these changes on treatment choices. Correlations between demographics and treatment choices were also assessed.

Qualitative data were derived from transcribed audio-recorded discussions with participants about reasons for their preferences and any changes thereto following the education session. Krueger's methodology for focus group analyses was used to categorize participants' reasons for preferences (Krueger and Casey, 2000; Onwuegbuzie *et al.*, 2009).

Results

Quantitative data

PARTICIPANTS BASELINE CHARACTERISTICS

In total, 24 of 32 (75%) participants were women with a mean (SD) age of 75 (6) years; 17 (53%) had a university degree (see Table 1).

All but 3 (29) participants knew of, or had cared for, someone who had dementia and 21 (66%) had a proxy decision maker in case they became incapable of treatment-related decision making. Of those caring for somebody with AD, approximately 10 were children (34%), 3 were a spouse or partner (10%), and the other 16 were more distant relatives or friends (55%). The mean (SD) scores on the Alzheimer's Knowledge Questionnaire were: 72% (14%) – a i.e. 7.2 questions correct out of 10 questions – before the education session; 78% (13%) immediately after; and 80% (18%) one month later (see Table 2). On the HBMQ, 30 (94%) participants rated developing AD as "very serious."

PARTICIPANTS' CHOICES BEFORE THE EDUCATION SESSION

Prior to the education session, there was a pattern of choosing less active treatments as AD progressed. Overall, the odds of preferring to receive a treatment decreased more than 50-fold when the condition progressed from "current health" (OR = 56.7) to "severe stage" (OR = 1) (see Table 3). In their current state of health, 86%

Table 2. Participant choices for treatment by AD severity and AD knowledge before and immediately after education session, and one month later

| TIME | CHOSEN TREATMENTS | AD SEVERITY | | | | MEAN AD KNOWLEDGE SCORE (SD) |
|---|---|-------------|-------|----------|--------|------------------------------|
| | | NONE | MILD | MODERATE | SEVERE | |
| Before education session | CPR and ventilation | 87.5% | 68.8% | 56.3% | 12.5% | 72.7% (14.1%) |
| | Hospitalization for tests and treatment | 93.8% | 81.3% | 62.5% | 31.3% | |
| | Relief of pain and symptoms at home | 93.8% | 90.6% | 83.9% | 59.4% | |
| Immediately after education session | CPR and ventilation | 87.5% | 68.8% | 50.0% | 9.4% | 78.7% (12.5%) |
| | Hospitalization for tests and treatment | 96.9% | 87.5% | 59.4% | 21.9% | |
| | Relief of pain and symptoms at home | 96.9% | 93.8% | 78.1% | 50.0% | |
| One month after education session (3 participants did not provide data) | CPR and ventilation | 81.5% | 69.2% | 32.0% | 3.7% | 83.0% (9.1%) |
| | Hospitalization for tests and treatment | 96.3% | 88.9% | 48.0% | 14.3% | |
| | Relief of pain and symptoms at home | 93.1% | 93.1% | 76.9% | 55.6% | |

of the participants would want to receive CPR, while only 13% would if they had severe dementia.

Those who were not close to someone with AD were much more likely (OR: 6.4) to want active treatment at the moderate and severe stages than those with direct experience with someone with AD. Being born in Canada, having a proxy, or a university education was associated with about half the odds of preferring to receive treatment compared to those who had none of these characteristics (see Table 3).

EFFECT OF THE EDUCATION SESSION ON PARTICIPANTS' CHOICES

There was a pattern of choosing less active treatments for the moderate and severe stages of AD after the education session. For example, 56% of the participants chose CPR for the moderate stage before the session, as compared to 32% at one month follow up. Similarly, 31% chose hospitalization for a medical issue during the severe stage of AD before the session, and 14% did at one month follow up. However, there was no statistically significant relationship between improving knowledge about AD with the education session and a change in preferences.

BPSD

In total, 12 (38%) and 10 (31%) participants reported that aggression or psychosis, respectively,

would be most worrisome among the five BPSDs examined (i.e. disinhibition, aggression, apathy, mood changes, and psychosis). No participants chose apathy as their most worrisome symptom.

Participants were asked to prioritize the use of psychotropic medications, physical restraints, and seclusion in terms of which measure they would prefer when managing the symptom they considered most worrisome. Thirty-one (97%) participants chose medications to manage BPSD. Only one participant chose physical restraints and no one chose seclusion as a first choice.

Qualitative data

REASONS FOR CHANGING PREFERENCES AFTER THE EDUCATION SESSION

The reasons for preferring less intensive interventions included gaining new information about AD or a particular symptom, envisioning family burdens becoming too great ("I don't want to be a burden on anyone," S1), or realizing that available medical interventions were futile, ("It looks like a futile effort to take the person to a hospital" S2) or considering life at that AD stage to be purposeless ["...with that (lack of self-care) coming into play, forget it," S2].

Unexpectedly, some participants' preferences for active treatment increased, rather than decreased,

Table 3. Coefficient estimates for the variables in the final model predicting treatment choices for Alzheimer's disease

| PARAMETER | ESTIMATE | SE | UPPER 95% CL | LOWER 95% CL | Z | P-VALUE | OR |
|-----------------------------|----------|--------|-----------------|-----------------|-------|---------|-------|
| Intercept | 1.1555 | 1.1189 | -1.0376 | 3.3486 | 1.03 | 0.3018 | 3.18 |
| Treat. - cardio. | -2.1281 | 0.4834 | -3.0754 | -1.1807 | -4.4 | <0.0001 | 0.12 |
| Treat. - hospital. | -1.1325 | 0.5419 | -2.1946 | -0.0703 | -2.09 | 0.0367 | 0.32 |
| Treat. - pain relief (ref.) | 0 | 0 | 0 | 0 | - | - | 1.00 |
| Sev. - as Is | 4.0551 | 0.6747 | 2.7328 | 5.3775 | 6.01 | <0.0001 | 57.69 |
| Sev. - mild | 2.9322 | 0.436 | 2.0777 | 3.7866 | 6.73 | <0.0001 | 18.77 |
| Sev. - moderate | 1.602 | 0.3252 | 0.9646 | 2.2393 | 4.93 | <0.0001 | 4.96 |
| Sev. - severe (ref.) | 0 | 0 | 0 | 0 | - | - | 1.00 |
| Pre-treatment | 0.3516 | 0.2576 | -0.1533 | 0.8566 | 1.36 | 0.1723 | 1.42 |
| Post-treatment | 0.268 | 0.249 | -0.2201 | 0.7562 | 1.08 | 0.2818 | 1.31 |
| Follow_up (ref.) | 0 | 0 | 0 | 0 | - | - | 1.00 |
| Knowledge | -0.8347 | 1.1519 | -3.0924 | 1.423 | -0.72 | 0.4687 | 0.43 |
| Female | 0.5581 | 0.3749 | -0.1767 | 1.2928 | 1.49 | 0.1366 | 1.75 |
| Age <70 | 0.7683 | 0.3994 | -0.0145 | 1.551 | 1.92 | 0.0544 | 2.16 |
| Age 71-79 | -0.1125 | 0.3639 | -0.8256 | 0.6007 | -0.31 | 0.7573 | 0.89 |
| Age 80+ | 0 | 0 | 0 | 0 | - | - | 1.00 |
| Born in Canada | -0.7602 | 0.303 | -1.354 | -0.1664 | -2.51 | 0.0121 | 0.47 |
| Proxy | -0.9197 | 0.3533 | -1.6121 | -0.2273 | -2.6 | 0.0092 | 0.40 |
| University | -0.8446 | 0.3245 | -1.4805 | -0.2086 | -2.6 | 0.0092 | 0.43 |
| Do not know Alzheimer | 1.8559 | 0.5934 | 0.693 | 3.0189 | 3.13 | 0.0018 | 6.40 |

Treat.: treatment; Sev.: severe; Cardio.: cardiopulmonary resuscitation.

after the education session. Reasons for preferring more active treatment included hope being ever present and life still mattering ("Because life is sweet. You don't really want to die," S3). These participants also explained these changes in terms of re-evaluating AD stages to be "not as bad as I thought," renewing a wish not to die, "When I listened to your explanation of moderate, then I thought 'moderate' is not too bad," S3 and re-embracing hope for some kind of recovery or new treatment ("Initially, I thought if I had severe, I don't think I'd really want to live. And then when I thought about it again, I thought there might be hope somewhere. And so I just changed my mind," S3).

Hospitalization was favoured in many cases because participants believed contemporary large, acute care hospitals had more options to improve the situation. Confidence in medical advances also explained preferences for hospitalization ("I think because the way research is going, they're going to come up with something somewhere along the line, not in the very near future, but it's like all other disease, they do come up with something eventually that helps," S3). Moreover, some participants believed hospitalization would result in fewer people being harmed than if the person remained in the home or long-term care facility.

REASONS FOR MOST WORRISOME SYMPTOM

Aggression was chosen as the most worrisome of the five BPSD symptoms because of its potential consequences for others ("I'm conscious of the fact that I'm a big guy and I could hurt someone. The important thing to me in my current state would be not to cause injury or damage to someone else," S4) and as being antithetical to personal self-image ("Because I mean all my life I've been a pacifist," S5). Reasons for choosing psychosis included the intolerability of being disconnected from one's surroundings ("Not to be in the real world. You're out of touch with reality," S6), increased risk to being harmful to others, and increased risk of having the other symptoms.

REASONS FOR MANAGEMENT OF BPSD

Reasons for the almost universal preference for medications focused on a perceived acceptable benefit-to-risk ratio, i.e. modest symptom relief and small probability of stroke or early death ("I'm prepared to gamble on the drugs...the lesser of two evils," S7). Reasons for preferring physical restraints over seclusion centered on being able to see and speak with other people nearby (i.e. healthcare staff). Reasons for preferring seclusion over physical restraints emphasized not being afraid of being alone ("I don't mind being alone. As a matter of fact, I prefer being alone," S8).

Furthermore, restraints were considered to be not just more restrictive, but for some participants, unimaginable and intolerable ("I've been fiercely independent my entire life and I think that's just a horror to me," S9). One participant described restraints as an anathema based on membership in a historically oppressed group.

Without solicitation or prompting by the researchers during the group discussions, a handful of participants either stated they would access physician assisted dying (PAD) if they developed AD or expressed hope that PAD would become legally permissible in the near future.

Discussion

We conducted a mixed-method study to assess the knowledge of older persons without dementia about AD and their choices for general medical interventions at various stages of AD and for BPSD. We then assessed whether a 20-min education session would change these choices. Participants perceived AD as a severe illness and they already had high baseline knowledge of AD. They chose less invasive options as AD progresses. Only about half of the participants chose pain relief in the severe stage of AD which was a counter-intuitive finding. A possible explanation comes from the focus groups where people responded that they would "just want to die" at this point in the illness and they did not want any kind of intervention. Not having a close friend or relative with dementia, having below university education, being born outside of Canada, and not having a proxy for decision making were all significantly associated with choosing more active options such as CPR and hospitalization in the later stages of AD. Becoming aggressive or psychotic during the course of AD was worrisome for participants and managing these symptoms with medications was preferred to seclusion or physical restraints, despite the potential adverse effects of medications.

This genesis of this study came from clinical practice. An elderly gentleman with severe dementia was admitted to a geriatric psychiatry unit because his escalating BPSD proved too difficult to manage in a long-term care home. His attending physician (REW) witnessed his devoted family's ambivalence as to whether the short-term benefits of repeated transfer to an emergency department for treatment of his condition made sense. Before developing his dementia, he had commented to family members that acute care hospitalization for advanced disease seemed ill-advised. Yet, there was no written or clearly expressed advance directive

that confirmed the strength and consistency of his views vis-à-vis dementia and more specifically about BPSD.

The hospital clinical ethicist (BJR) recommended caution, given that some studies show that neither relatives nor physician beliefs about patient preferences are accurate, despite their loyalty to the patient. The person himself would be the most appropriate evaluator of symptoms such as psychosis and apathy. It is, however, not enough to know which symptoms he – when capable – would have considered tolerable versus intolerable. Knowing why (i.e. what is at stake) is ethically important: for instance, do the symptoms make daily activities more exhausting, less enjoyable, or highly distressing? Do they contradict one's character or identity? Or perhaps they erode personal dignity?

This study highlighted the potential value of discussing advance directives with patients with respect to developing AD specifically. Educational AD and BPSD tools need to be developed with physicians and families in mind. Community physicians' recommendations to their patients and families to use publicly available tools should be influential. At the same time, families must be supported to understand what lies ahead when a loved one is first diagnosed with AD, both for their own well-being as well as the patient's well-being.

This study has several limitations. First, there was no control group, therefore determining the impact of education on treatment choices relied on a significant change in knowledge of AD prior to and after the education session. This was challenging because there was a relatively low number of participants and their experience with AD and high knowledge at baseline may have limited our ability to detect statistically significant changes and predictors for these changes. Using the complete Alzheimer's Disease Knowledge Scale (Carpenter *et al.*, 2009) may help to better capture knowledge base and changes. Second, there was likely a sample bias given that several participants appeared to have volunteered because they were seeking out or confirming information about AD. Future studies should recruit seniors who are not as self-motivated to learn about AD and religion's impact on end of life decision making should be studied to help ensure culturally sensitive directives are developed. Finally, because seniors with mild or newly diagnosed dementia may have been alarmed or frightened to learn for the first time about BPSD symptoms, we excluded this population. Future studies should recruit these seniors because they have the most immediate need for advance care planning for dementia and BPSD and are likely still capable to share their own

priorities, values, and reasons for and against care options.

Conclusions

The results of our study support the value of a larger study to better understand seniors' choices, decisions, and reasons for preferring treatment and care options for AD and associated BPSD. Some studies show that video can be an effective educational tool (Volandes *et al.*, 2007). A future study could compare the impact and accessibility of brief video portrayals of AD stages and BPSD symptoms with brief verbal descriptions on participants' attitudes and treatment preferences. Future studies should consider conducting individual interviews rather than using a group format.

Legislation allowing PAD for people with certain health conditions in Belgium, the Netherlands, and Canada, and related proposals in the United States, are generating much professional and organizational discussion. Relevant to this pilot study, it is critically important to empower people to explain to their families and clinicians what an illness like AD signifies to them and how medical interventions do or do not align with preserving what is meaningful to them in terms of personal identity, treasured relationships and responsibilities, and quality of life in light of the human reality of declining health and foreseeable death. Advance directives, and the reflections and discussions they hopefully represent, are becoming even more important mechanisms to support relational autonomy, interdependence, and solidarity.

Acknowledgments

The authors are grateful to the contributions of each study participant, Tamara Arénovich, Dielle Miranda, and audiences of our presentations about this study. This work was supported by the Physicians' Services Incorporated (PSI) Foundation.

All authors meet the authorship criteria according to the latest guidelines of the International Committee of Medical Journal Editors. Drs. Waxman, Mulsant and Russell were responsible for the study conception and design. Mr. Iu performed data analysis and Drs. Waxman and Russell drafted the manuscript. All authors critically reviewed the manuscript and accepted the final version. All authors agree to be accountable for all aspects of the work. This manuscript is original research, which has not been published, nor is under consideration for publication elsewhere.

Supplementary material

To view supplementary material for this article, please visit <https://doi.org/10.1017/S1041610218000479>

References

- Aleccia, J.** (2017). *End-of-life Advice: More than 500,000 Chat on Medicare's Dime*. Kaiser Health News. 14 August 2017. Available at: <https://khn.org/news/end-of-life-advice-more-than-500000-chat-on-medicare-dime/>; last accessed 22 August 2017.
- Arai, Y., Arai, A. and Zarit, S.** (2008). What do we know about dementia?: a survey on knowledge about dementia in the general public of Japan. *International Journal of Geriatric Psychiatry*, 23, 433–438.
- Boustani, M. et al.** (2008). Measuring primary care patients' attitudes about dementia screening. *International Journal of Geriatric Psychiatry*, 23, 812–820.
- Carpenter, B. et al.** (2009). The Alzheimer's disease knowledge scale: development and psychometric properties. *Gerontologist*, 49, 236–247.
- Deep, K. et al.** (2010). "It helps me see with my heart": how video informs patients' rationale for decisions about future care in advanced dementia. *Patient Education and Counseling*, 81, 229–234.
- Fried, T. R. et al.** (2011). Agreement between older persons and their surrogate decision-makers regarding participation in advance care planning. *Journal of American Geriatrics Society*, 59, 1105–1109.
- Gjerdingen, D. et al.** (1999). Older persons' opinions about life-sustaining procedures in the face of dementia. *Archives of Family Medicine*, 8, 421–425.
- Hamann, J. et al.** (2010). Can psychiatrists and neurologists predict their patients' participation preferences? *Journal of Nervous and Mental Disease*, 198, 309–311.
- Hersch, E. C. and Falzgraf, S.** (2007). Management of the behavioral and psychological symptoms of dementia. *Clinical Interventions in Aging*, 2, 611–621.
- Janz, N. K. and Becker, M. H.** (1984). Health belief model: a decade later. *Health Education Quarterly*, 11, 1–47.
- Kim, H. M., Chiang, C. and Kales, H. C.** (2011). After the black box warning: predictors of psychotropic treatment choices for older patients with dementia. *Psychiatric Services*, 62, 1207–1214.
- Kolanowski, A. et al.** (2010). It's about time: use of nonpharmacological interventions in the nursing home. *Journal of Nursing Scholarship*, 42, 214–222.
- Krueger, R. A. and Casey, M. A.** (2000). *Focus Groups: A Practical Guide for Applied Research*. Thousand Oaks: Sage Publications.
- Leone, E. et al.** (2009). Methodological issues in the non-pharmacological treatment of BPSD in nursing home—the TNM study. *Journal of Nutrition Health and Ageing*, 13, 260–263.
- Lyketsos, C. B. et al.** (2002). Prevalence of neuropsychiatric symptoms in dementia and mild cognitive impairment: results from the cardiovascular health study. *JAMA*, 288, 1475–1483.

- Madhusoodanan, S.** (2001). Introduction: antipsychotic treatment of behavioural and psychological symptoms of dementia in geropsychiatric patients. *American Journal of Geriatric Psychiatry*, 9, 283–288.
- Margallo-Lana, M. et al.** (2001). Prevalence and pharmacological management of behavioural and psychological symptoms amongst dementia sufferers living in care environments. *International Journal of Geriatric Psychiatry*, 16, 39–44.
- Morrison, R. S. and Siu, A. L.** (2000). Survival in end-stage dementia following acute illness. *JAMA*, 284, 47–52.
- Narang, P. et al.** (2010). Antipsychotic drugs: sudden cardiac death among elderly patients. *Psychiatry*, 7, 25–29.
- National Cancer Institute** (2011). *Theory at a Glance: A Guide for Health Promotion Practice: Health Belief Model*. Available at: <http://www.cancer.gov/cancertopics/cancerlibrary/theory.pdf>; last accessed November 22, 2011.
- Onwuegbuzie, A. J. et al.** (2009). A qualitative framework for collecting and analyzing data in focus group research. *International Journal of Qualitative Methods*, 8, 1–21.
- Rurup, M. L. et al.** (2005). Physicians' experiences with demented patients with advance euthanasia directives in the Netherlands. *Journal of American Geriatric Society*, 53, 1138–1144.
- Shalowitz, D. I., Garrett-Mayer, E. and Wendler, D.** (2006). The accuracy of surrogate decision makers: a systematic review. *Archives of Internal Medicine*, 166, 493–497.
- Stevens, J. R. et al.** (2014). Strategies for the prescription of psychotropic drugs with black box warnings. *Psychosomatics*, 55, 123–133.
- Thies, W and Bleil, L.** (2013). Alzheimer's disease facts and figures. *Alzheimer's & Dementia*, 9, 208–245.
- Uhlmann, R. F., Pearlman, R. A. and Cain, K. C.** (1988). Physician' and spouses' predictions of elderly patients' resuscitation preferences. *Journal of Gerontology*, 43, M115–121.
- Volandes, A. E. et al.** (2007). Using video images of dementia in advance care planning. *Archives of Internal Medicine*, 167, 828–833.
- Yung, V. Y. et al.** (2010). Documentation of advance care planning for community-dwelling elders. *Journal of Palliative Medicine*, 13, 861–867.



International Psychogeriatric Association
Better Mental Health for Older People

Submit an Article for the IPA Bulletin!

The *IPA Bulletin* is the quarterly newsletter of the International Psychogeriatric Association (IPA). In addition to news about IPA and related organizations, the *IPA Bulletin* contains articles on recent advances and initiatives in the field in of psychogeriatrics. The *IPA Bulletin* reaches all the members of IPA, including our affiliates around the world.

Research and Practice. The Research and Practice section of the *IPA Bulletin* highlights recent advances in psychogeriatrics and related fields. Articles from all fields related to the mental health of older people are welcome.

Around the World. The Around the World section of the *IPA Bulletin* features articles by clinicians and researchers which report on activities and issues within their country or region. These articles seek to educate others on the state of the field in other countries and provide unique perspectives from around the world.

To learn more about the *IPA Bulletin*,
visit our website:

www.ipa-online.org

LETTER TO THE EDITOR

doi:10.1017/S1041610218000248

Introducing a family intervention to elderly with first episode psychosis

Schizophrenia guidelines list family interventions as an efficient means in reducing relapses. Interventions aim to help families cope with their relative's problems more effectively, provide support and education, and reduce levels of distress and improve the family communication (see deHaan *et al.*, 2002).

There are only a few studies available on three-generation mental health, mostly affective disorders, and hardly any including four generations. Including the worries of underage children in the interventions is an important aspect of family psychoeducation and a preventive measure in mental health (Solantaus *et al.*, 2010).

Our study is a sub-study of the Helsinki Old-Age Psychosis Study (HOPS) (Louhija *et al.*, 2017). We aimed at inventorying the needs of families of hospitalized first episode psychosis (FEP) persons over 60 years of age. If the close family included underage children, the patient and the family were offered a brief modeled psychoeducational intervention named “Let's talk about grandchildren” (Solantaus *et al.*, 2010). Out of the five sessions, one was held separately for the patient, two for the family members, and two for the patient and the family together. The families were encouraged to talk about grandparent illness, current problems, and answer children's questions. Psychiatric nurses trained for psychoeducation were responsible for the intervention. The patient's nursing care managers participated in sessions.

A revised version of the Care Burden Scale for relatives (Bergmark and Wistedt, 1989) was applied. The participants evaluated the intervention by a 15-item visual analogue scale.

In total, 17 out the first 50 HOPS-participants met the inclusion criteria. Eight families agreed to participate and four completed the program. Two patients withdraw for medical reasons and two for other reasons. All four patients were females. The family members were one spouse, two daughters, and two sons.

The families had experienced a change in life routines and distress caused by the unpredictability of the patient's behavior. Psychotic behavior caused challenges especially when the patient had no insight about her condition. Risk of inheritance

was brought up. No children attended but grandmother's behavior was discussed at home.

The family members felt that the intervention gave them “more words” to discuss the illness. Information concerning patient's mental condition and psychiatric treatment was valued. All families had a positive attitude to medication. The evaluations given by the patients were rather neutral: “it is useful to meet with the experts.”

The reluctance of the elderly to reveal about “personal” matters may partly explain the low recruiting level. Family psychoeducation may be most suitable when the need for information is greatest. If introduced too early, the patient may not be able to discuss any family issues; if introduced close to discharge, the patient may not be motivated to any new interventions.

Individual customization and sessions tailored according to one's needs are necessary. The intervention should be integrated to the patient's psychiatric treatment. Intervention approaches suiting the needs of families not used to psychosocial ways of thinking are needed (Leavey *et al.*, 2004) as well as studies looking at the effectiveness of the psychoeducation with FEP patients.

Conflict of interest

None.

Description of authors' roles

T. Saarela formulated the research question, designed the study, coordinated the data collection, analyzed the data, and had main responsibility for writing the article. U. Louhija supervised the identification and selection of the participants and assisted with writing the article. M. Johansson planned and supervised the family interventions and participated in data analysis. B. Appelberg assisted with participant selection, supervised the research process, and assisted with writing the article. K. Juva assisted with participant selection and writing the article.

Acknowledgments

We thank all families who participated in this study and our family therapists Jasmi Jauhonen and Riitta Alopaeus for their contribution.

References

- Bergmark, T. and Wistedt, B.** (1989). CBS-R: a new scale for burden on relatives, and effects of education. Unpublished report. Psykiatriska Institutionen, Danderyds Sjukhus.
- deHaan, L. et al.** (2002). Priorities and satisfaction on the help needed and provided in a first episode psychosis. A survey in five European Family Associations. *European Psychiatry*, 17, 425–433.
- Leavey, G., Gulamhussein, S., Papadopoulos, C., Johnson-Sabine, E., Blizard, B. and King, M.** (2004). A randomized controlled trial of a brief intervention for families of patients with a first episode of psychosis. *Psychological Medicine*, 34, 423–431.
- Louhija, U. M., Saarela, T., Juva, K. and Appelberg, B.** (2017). Brain atrophy in first episode psychosis. *International Psychogeriatrics*, 29, 1925–1929. doi:10.1017/S1041610217000953.
- Solantaus, T., Paavonen, E. J., Toikka, S. and Punamäki, R. L.** (2010). Preventive interventions in families with parental depression: children's psychosocial symptoms and prosocial behavior. *European Child & Adolescent Psychiatry*, 19, 883–892. doi:10.1007/s00787-010-0135-3.

TUULA SAARELA,¹ MONICA JOHANSSON,²
ULLAMARJA LOUHIJA,³ BJÖRN APPELBERG¹
AND KATI JUVA³

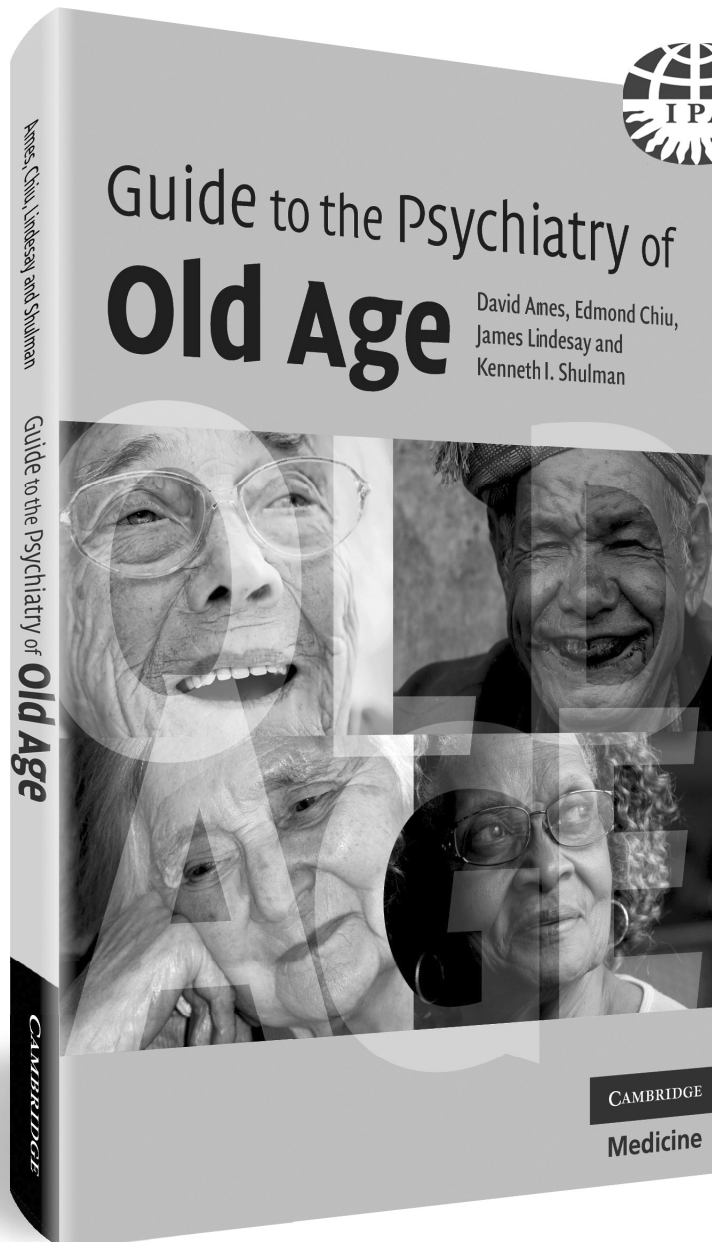
¹Department of Psychiatry, Helsinki University Hospital, Helsinki, Finland

²Preventive and Psychoeducational Family Interventions, Functional Child & Family Methods, Heinola, Finland

³Department of Geriatric Psychiatry, Helsinki University Hospital, Helsinki, Finland
Email: tuula.saarela@kolumbus.fi

CAMBRIDGE

Medicine



Endorsed by the International
Psychogeriatric Association

David Ames, *University of Melbourne*
Edmond Chiu, *University of Melbourne*
James Lindsay, *University of Leicester*
Kenneth Shulman, *University of Toronto*

**25% discount
for IPA Members**

Enter promotional code
IPA10 when you add to basket

order here
www.cambridge.org/9780521681919

Paperback 9780521681919

July 2010
GBP 29.99

Compact, accessible and affordable

Each chapter is sharp, clear and practical, enhanced by tables and diagrams for quick assimilation and reference on the ward or in the clinic

Coverage also includes legal and ethical issues, and the neglected topic of alcohol and drug abuse in the elderly

www.cambridge.org/medicine



CAMBRIDGE
UNIVERSITY PRESS



International Psychogeriatric Association

Better Mental Health for Older People

IPA NEEDS YOU!



About IPA

The International Psychogeriatric Association (IPA), founded in 1982 and representing members in more than 50 countries, is a unique and diverse professional healthcare community promoting better geriatric mental health – across disciplines, across borders, and across geriatric issues. Psychiatrists, scientists, neurologists, geriatricians, primary care physicians, epidemiologists, nurses, psychologists, occupational therapists, social workers, and many other healthcare professionals come to the IPA community from all around the world to discuss, learn, share and research information about behavioral and biological aspects of geriatric mental health. IPA promotes research and education, facilitates an international exchange of ideas, and fosters cross-cultural understanding of the latest developments in the field.

IPA Membership

The International Psychogeriatric Association (IPA) encourages professionals from all backgrounds related to geriatric mental health to join our healthcare community. Here are some of the many benefits of joining IPA:

- Unique opportunities to interact with colleagues around the world how share an interest in advancing research, education, and theory about mental health care in older adults.
- Valuable discounts on participation in education and scientific meetings which reflect the full spectrum of disciplines related to psychogeriatrics. Reduced registration rates for the annual IPA International Congress and IPA Regional Meetings are included.
- Relevant publication on updates in the field, including *International Psychogeriatrics*, IPA's peer reviewed journal which publishes twelve issues per year and additional special-focus supplements.
- Access to online educational materials and *The IPA Complete Guides to Behavioral and Psychological Symptoms of Dementia (BPSD)*.
- Subscription to the *IPA Bulletin*, IPA's newsletter which features articles on advances in the field and information on upcoming programs around the world.
- Participation in the IPA Member Forums – smaller member groups which enable members to connect with one another and collaborate on research and clinical care practices. These forums reflect the diversity of interests within geriatric mental health.

How to Join

To learn more about IPA and become a member, please visit our website: www.ipa-online.org. IPA offers a one-year membership for \$160 USD. One-year student or retired memberships are available for \$99 USD. **A limited number of Lifetime Memberships are also available for \$1000 USD.** The IPA Secretariat is available to answer any questions you may have about IPA or membership. Please contact us at +1 414 918 9889 or by email at info@ipa-online.org.

Have Questions? Contact us!

International Psychogeriatric Association
 555 East Wells Street, Suite 1100
 Milwaukee, WI 53202
 United States
 Phone: +1 414 918 9889
 Fax: +1 414 276 3349
 Twitter: @IPA_Online
www.ipa-online.org
info@ipa-online.org

The collage features three main components:

- IPA Bulletin:** A newsletter cover with the title 'IPA Bulletin' and the subtitle 'INTERNATIONAL PSYCHOGERIATRIC ASSOCIATION • BETTER MENTAL HEALTH FOR OLDER PEOPLE'. It includes a 'WHAT'S INSIDE' section and a 'PRESIDENT'S MESSAGE' by Raimundo Mateos.
- International Psychogeriatrics:** The cover of the peer-reviewed journal, featuring the title 'International Psychogeriatrics' and the subtitle 'Psychogeriatrics English'.
- IPA Complete Guides to Behavioral and Psychological Symptoms of Dementia (BPSD) Specialists Guide:** A guide cover with the title 'The IPA Complete Guides to Behavioral and Psychological Symptoms of Dementia' and a sub-section for 'BPSD Specialists Guide'. It features several small portraits of individuals.