

## COMMENTARY

# Role of behavioral problems in carer burden in young-onset dementia

Commentary on “Carer burden and behavioural disturbance is similar between younger-onset Alzheimer’s Disease and behavioural-variant frontotemporal dementia” by Kang *et al.*

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Family carers of people with dementia are more likely to experience moderate to high carer burden (Cullinan *et al.*, 2021) and carers of people with young-onset dementia (YOD) defined as dementia with symptom onset before 65 years of age (Rossor *et al.*, 2010), face additional challenges. Younger patients tend to have a faster disease progression with more atypical presentations, including a higher prevalence of language impairment, agnosia, and behavioral involvement (Mendez, 2006). Community support for these young patients tends to be minimal and the loss of employment due to dementia results in a greater socioeconomic burden (Kandiah *et al.*, 2016). Dementia in the young have a higher genetic susceptibility and positive family history would also affect the carer burden in view of the previous negative experience with dementia outcomes (Rossor *et al.*, 2010).

Carer burden has been associated with factors related to the clinical features of the patient including presence of behavioral and psychological disorders (BPSD) (Feast *et al.*, 2016; Liu *et al.*, 2018). Carers of people with YOD suffer high levels of burden but the difference when compared to late-onset dementia is not conclusive (van Vliet *et al.*, 2010). A study that evaluated 183 patient-carer dyads to compare carer burden in YOD ( $n=57$ ) with late-onset dementia ( $n=126$ ) found higher levels of carer burden in YOD (Lim *et al.*, 2018). They reported that risk factors for high caregiver burden in YOD included family history of dementia and behavioral symptoms, which included disinhibited behavior, delusions, and apathy as measured by neuropsychiatric inventory-questionnaire (Cummings *et al.*, 1994; Lim *et al.*, 2018).

Few have investigated the role of individual symptoms of BPSD in impacting on carer’s burden. A systematic review, which included forty medium and high quality quantitative papers, showed depressive behaviors were the most distressing for caregivers followed by agitation/aggression and apathy (Feast *et al.*, 2016). The authors had included “aged” in the key terms for search and hence included studies mostly with older adults (Feast *et al.*, 2016). In a study wherein 92 consecutive caregivers of YOD were recruited from the Cognitive Neurology Clinics of Modena, Northern Italy, BPSD was one of the determinants of carer burden (total NPI score), however, individual NPI domains were not examined (Chiari *et al.*, 2021). In another study that used Zarit Burden Inventory (Zarit *et al.*, 1987), caregiver burden was measured for 3 main clinical variants of Frontotemporal dementia (FTD) — behavioral variant frontotemporal dementia (bvFTD,  $n=17$ ), semantic dementia (SemDem,  $n=20$ ), and progressive nonfluent aphasia (PNFA bvFTD,  $n=20$ ), and Alzheimer disease (AD,  $n=19$ ) patients (Mioshi *et al.*, 2013). Caregivers of bvFTD patients reported the highest burden, whereas SemDem and PNFA caregivers reported burden similar to AD (Mioshi *et al.*, 2013). Behavioral changes in FTD were strongly associated with caregiver burden, however, it seemed that a combination of functional impairment and behavioral changes, rather than behavioral dysfunction alone, were the main predictors of caregiver burden (Mioshi *et al.*, 2013).

Most studies have reported on carer burden in dementia due to BPSD in the elderly and few studies have focussed and specifically compared between different YOD subtypes. Kang *et al.* (2023) in a

recent paper in *International Psychogeriatrics*, addressed these aspects and the results presented are significant as the authors compared carer burden between young-onset AD and behavioral variant frontotemporal dementia (bvFTD) for their associations with clinical correlates such as behavior problems, daily skills, and cognition. Specifically, Kang *et al.* (2023) carried out retrospective cross-sectional study of patient-carer dyads with YOD who attended a tertiary service over a five-year period and were part of a larger study *Biomarkers in younger-onset neurocognitive disorders* (BeYOND study) (Loi *et al.*, 2021). Of the 33 carers ( $n = 20$  with AD and  $n = 13$  with bvFTD) whose data was included, majority were females (21 females, 68%) and there were no differences in their demographics. For the patients, cognition was evaluated with Neuropsychiatry Unit Cognitive Assessment tool (NUCOG) (Walterfang *et al.*, 2006) and behavioral changes with the Cambridge Behavioral Inventory-Revised (CBI-R) (Wear *et al.*, 2008). Zarit Burden Inventory-short version (Bédard *et al.*, 2001) was used to measure carer burden. The mean total NUCOG score overall was 63.6 ( $SD \pm 16.7$ ) indicating moderate cognitive impairment. There were no differences in domains such as memory, orientation, duration of symptoms, and caregiving period between the two groups. Compared to patients with AD, patients with bvFTD were more likely to have difficulty with stereotypic and motor behaviors. Patients with AD were found to have difficulties with everyday skills. Carer burdens were high but did not differ significantly between AD (55%, 11/20; mean = 18.5) and bvFTD (38%, 5/13; mean = 15.2). The major findings of Kang *et al.* (2023) are that although they found overall behavioral disturbance, as well as mood and belief related, were significantly associated with carer burden in YOD, they did not find any difference in carer burden between young-onset AD and bvFTD groups. Unexpectedly they failed to find differences in behavioral disturbance between the two subtypes, which may explain the similar levels of carer burden in the two groups. Kang *et al.* (2023) highlight the importance of clinicians routinely screening for behavioral disturbance in younger-onset dementia, irrespective of presumed pathological diagnosis. They also emphasize assessments for BPSD in the initial phase with standardized measures. This is important especially in light of recent report of greater caregiver burden in people with apathy in mild cognitive impairment (MCI) (Connors *et al.*, 2023). Presence of mild behavioral impairment (MBI) in patients with mild cognitive impairment or subjective cognitive decline MCI has also been associated with greater carer burden, with each of the five MBI domains

(affective dysregulation, impulse control, decreased motivation, social inappropriateness, and abnormal perception or thought content) showing significant association with carer burden (Sheikh *et al.*, 2018).

While the Kang *et al.* (2023) study expands on differences in carer burden in YOD subtypes such as young-onset AD and bvFTD, we must consider the strengths and limitations of the study. Kang *et al.* (2023) used retrospective data from patients who either attended as inpatient or outpatient from a single hospital in Australia. Although the findings may not be generalizable to other settings, they add to the growing literature indicating similar behavior impairment and carer burden in YOD subtypes. Kang *et al.* (2023) assessed cognition using the NUCOG and used Mini-Mental State Examination (Folstein *et al.*, 1975) for an “equivalent” total score where patients were unable to complete NUCOG, which is a crude cognitive measure and limited in examining different domains. Future studies incorporating comprehensive neuropsychological testing would help to examine level of cognitive impairment and specific cognitive domains associated with carer burden. The behavior disturbance items on the CBI-R used by Kang *et al.* (2023) do not measure different BPSD domains and sub-domains. CBI-R is a shorter version of CBI to identify disease-specific behavior patterns and includes cognition, everyday skills, and self-care as domains that should ideally be assessed separately from core behavioral problems. Ideally, behavior and daily activities skills rating scales should be able to measure different dimensions and be able to measure them independently, since they are known to be associated with carer burden as distinct factors (Feast *et al.*, 2016; Liu *et al.*, 2018). There is need to examine and better understand the relationship between individual BPSD and carer burden, as it is essential to determine allocation of resources and interventions for BPSD management to target these behaviors. Appropriate and timely interventions can help to ameliorate these effects and, in turn, improve the quality of life of both caregivers and patients.

Challenges with the lack of consistency in BPSD measurement (neuropsychiatric domains, frequency, or prevalence) and the measurement of caregiver well-being (the use of interchangeable terminology such as stress, strain, and burden meaning the same construct) are well known and limit the ability to reach a consensus as to which BPSD result in the most negative outcomes for caregivers (Feast *et al.*, 2016). Another important factor is caregiver characteristics which have been shown to account for twice the variance of person with dementia characteristics when reporting neuropsychiatric symptoms (Sink *et al.*, 2006). In

addition, caregiver characteristics such as gender, overload, role captivity, confidence, neuroticism, and relationship quality are significant predictors for variance in carer burden too (Campbell *et al.*, 2008). Hence, it is important that future studies should focus on identifying pathways for the association between individual BPSD, caregiver variables, and caregiver outcomes. It is also important to have measures that address the contextual and caregiver characteristics associated with the behavior reporting. Larger collaborative studies comprising younger subjects from different cultural and geographic regions will be also needed.

In summary, Kang *et al.* (2023) extend previous research by not only evaluating high carer burden in YOD but also comparing them between two subtypes i.e., bvFTD and young-onset AD. Kang *et al.* (2023) also established overall behavioral disturbance as well as mood and belief related were significantly associated with carer burden in YOD, making them an important factor for pharmacological and non-pharmacological interventions. It is important to identify the domains of BPSD from initial stages so patients and carers may benefit from appropriate and tailored interventions.

## Conflict of interest

None.

## References

- Bédard, M., Molloy, D. W., Squire, L., Dubois, S., Lever, J. A., & O'Donnell, M. (2001). The Zarit Burden interview: a new short version and screening version. *Gerontologist*, 41(5), 652–657.
- Campbell, P., Wright, J., Oyebode, J., Job, D., Crome, P., Bentham, P., Jones, L., & Lendon, C. (2008). Determinants of burden in those who care for someone with dementia. *Int J Geriatr Psychiatry*, 23(10), 1078–1085.
- Chiari, A., Pistoressi, B., Galli, C., Tondelli, M., Vinceti, G., Molinari, M. A., Addabbo, T., & Zamboni, G. (2021). Determinants of caregiver Burden in early-onset dementia. *Dement Geriatr Cogn Dis Extra*, 11(2), 189–197.
- Connors, M. H., Teixeira-Pinto, A., Ames, D., Woodward, M., & Brodaty, H. (2023). Apathy and depression in mild cognitive impairment: distinct longitudinal trajectories and clinical outcomes. *International Psychogeriatrics*, 35(11), 633–642.
- Cullinan, J., Fealy, G., Lafferty, A., O'Shea, E., & Teahan, Á. (2021). An analysis of carer burden among family carers of people with and without dementia in Ireland. *International Psychogeriatrics*, 33(4), 347–358.
- Cummings, J. L., Mega, M., Gray, K., Rosenberg-Thompson, S., Carusi, D. A., & Gornbein, J. (1994). The neuropsychiatric inventory: comprehensive assessment of psychopathology in dementia. *Neurology*, 44(12), 2308–2314.
- Feast, A., Moniz-Cook, E., Stoner, C., Charlesworth, G., & Orrell, M. (2016). A systematic review of the relationship between behavioral and psychological symptoms (BPSD) and caregiver well-being. *International Psychogeriatrics*, 28(11), 1761–1774.
- Folstein, M. F., Folstein, S. E., & 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.
- Kandiah, N., Wang, V., Lin, X., Nyu, M. M., Lim, L., Ng, A., Hameed, S., & Wee, H. L. (2016). Cost related to dementia in the young and the impact of etiological subtype on cost. *Journal of Alzheimers Disease*, 49(2), 277–285.
- Kang, M. J. Y., Farrand, S., Evans, A., Chiu, W.-H., Eratne, D., Kelso, W., Walterfang, M., Velakoulis, D., & Loi, S. M. (2023). Carer burden and behavioral disturbance is similar between younger-onset Alzheimer's disease and behavioral variant frontotemporal dementia. *International Psychogeriatrics*, 1–8. <https://doi.org/10.1017/S1041610222001259>
- Lim, L., Zhang, A., Lim, L., Choong, T.-M., Silva, E., Ng, A., Kandiah, N., & Grill, J. (2018). High caregiver Burden in young onset dementia: what factors need attention? *Journal of Alzheimers Disease*, 61(2), 537–543.
- Liu, S., Liu, J., Wang, X.-D., Shi, Z., Zhou, Y., Li, J., Yu, T., & Ji, Y. (2018). Caregiver burden, sleep quality, depression, and anxiety in dementia caregivers: a comparison of frontotemporal lobar degeneration, dementia with Lewy bodies, and Alzheimer's disease. *International Psychogeriatrics*, 30(8), 1131–1138.
- Loi, S. M., Eratne, D., Cadwallader, C., & Ravanfar, P. (2021). Investigating the clinical utility of biomarkers and other novel tests in younger onset neurocognitive disorders: the BeYOND study, protocol for a longitudinal clinical study in a real-world setting. *Psychiatry and Clinical Psychology*, preprint. <https://doi.org/10.1101/2021.07.03.21259825>
- Mendez, M. F. (2006). The accurate diagnosis of early-onset dementia. *Int J Psychiatry Med*, 36(4), 401–412.
- Mioshi, E., Foxe, D., Leslie, F., Savage, S., Hsieh, S., Miller, L., Hodges, J. R., & Piguet, O. (2013). The impact of dementia severity on caregiver burden in frontotemporal dementia and alzheimer disease. *Alzheimer Dis Assoc Disord*, 27(1), 68–73.
- Rossor, M. N., Fox, N. C., Mummery, C. J., Schott, J. M., & Warren, J. D. (2010). The diagnosis of young-onset dementia. *Lancet Neurology*, 9(8), 793–806.
- Sheikh, F., Ismail, Z., Mortby, M. E., Barber, P., Cieslak, A., Fischer, K., Granger, R., Hogan, D. B., Mackie, A., Maxwell, C. J., Menon, B., Mueller, P., Patry, D., Pearson, D., Quickfall, J., Sajobi, T., Tse, E., Wang, M., Smith, E. E., & for the PROMPT registry investigators (2018). Prevalence of mild behavioral impairment in mild cognitive impairment and subjective cognitive decline, and its association with caregiver Burden. *International Psychogeriatrics*, 30(2), 233–244.
- Sink, K. M., Covinsky, K. E., Barnes, D. E., Newcomer, R. J., & Yaffe, K. (2006). Caregiver characteristics are

associated with neuropsychiatric symptoms of dementia.

*Journal of The American Geriatrics Society*, 54(5), 796–803.

**van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., & Verhey, F. R.** (2010). Impact of early onset dementia on caregivers: a review. *Int J Geriatr Psychiatry*, 25(11), 1091–1100.

**Walterfang, M., Siu, R., & Velakoulis, D.** (2006). The NUCOG: validity and reliability of a brief cognitive screening tool in neuropsychiatric patients. *Aust N Z J Psychiatry*, 40(11-12), 995–1002.

**Wear, H. J., Wedderburn, C. J., Mioshi, E., Williams-Gray, C. H., Mason, S. L., Barker, R. A., & Hodges, J. R.** (2008). The Cambridge Behavioural Inventory revised. *Dementia & Neuropsychologia*, 2, 102–107. <https://doi.org/10.1590/S1980-57642009DN20200005>

**Zarit, S. H., Anthony, C. R., & Boutsellis, M.** (1987).

Interventions with care givers of dementia patients: comparison of two approaches. *Psychology and Aging*, 2(3), 225–232.