Author's response to reviews

Title: Using U.S. Medicare Records to Evaluate the Indirect Health Effects on Spouses: a Case Study in Alzheimer's Disease Patients

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Dear BMC HSR:

Thank-you for giving us the opportunity to revise our manuscript, “Using Medicare Records to Evaluate the Health Effects of Spousal Caregiving for Alzheimer’s Disease,” in consideration of the reviewers’ comments. The revised manuscript is included in our uploaded submission, and the following pages provide our detailed response to the reviewers.

Sincerely,

Daniel Gilden,
President, JEN Associates
Dominique Somme:

This article reports an important and original approach of health consequences of a particular caregiving experience (the one of the spouses) of an AD patient. It is a very well written paper on very sound data, well criticized. The bibliography is appropriate and the length of the paper is adequate.

Nevertheless I have three major compulsory revisions:

1. The specificity of the AD versus all other possible chronic disease is not discussed. The methodology used selected the “control” group on demographic but not on the fact of being a spouse of a person suffering from a chronic disease. Indeed, in the control group the two “spouses” have very similar profile (Table 4, column 2 and 4). Thus it is impossible to conclude that it is “the AD diagnosis” that produces this impact on health care cost of the spouse rather than any other chronic disease that influence the health care cost of the index person (the difference is huge between column 2 and 3, table 4). In my point of view, this point has to appear in the discussion.

Authors’ response: Dr. Somme raises a very important issue. We did not compare different specific chronic diseases in this study. This would have required multiple stratifications by different chronic disease states but with little literature to guide which comorbidities or how many strata of such to use. Instead, we chose a very broad based algorithm employing almost all potential comorbidities (JEN Frailty Index) which was developed (and has been published) demonstrating effectiveness in analyses of economic endpoints. We have added the following paragraph to the discussion section (pages 16, lines 313-322) to help address this concern and justify our choice:

The question also arises as to whether spouses of patients with other chronic diseases would experience similar increases in healthcare costs. There are rare examples of this phenomenon existing for other disease states [52] or in association with widowhood from decedents of other chronic conditions [10]. The goal of our study was to find a connection between spouses’ healthcare status and their partners’ AD diagnosis while statistically controlling for a broad spectrum of impairments with the JFI. This temporal association is illustrated in Figure 1. One can speculate that AD’s particular behavioral and cognitive effects are especially stressful for spouses, both emotionally and in terms of caregiving burden. It will be a fruitful line of research to compare the spousal results
presented in Table 4 and Figure 1 with results obtained for other chronic diseases. The methods we establish here can form the basis for such research.

In addition, we have revised Table 4, which was confusing in its previous state. We eliminated the columns on AD patients and the spouses of the controls since these populations were not part of the study findings reported here. For consistency, we did the same to Table 3.

2. The reader does not have information on the heterogeneity of the groups. My hypothesis is that the variability in comorbidity and expenditures is more important in AD spouses than in “Spouses of matched control” (in relation with heterogeneity of coping) but I am very interested in having more clear idea on that particular point.

Authors’ response: The revised version of Table 4 includes additional cost information, with 95% CIs. This table shows that mean total yearly Medicare expenditures greatly exceed medians indicating that high-cost outliers have considerably affected the mean. At the same time, the 95% CIs were relatively narrow in both the AD spouses and the matched controls. This observation suggests that the difference in mean healthcare expenditures was due to a generalized elevation of costs among the AD spouses and not just a shift among high-cost outliers.

The regression analysis in Table 4 was constructed in stepwise fashion. It attempted to adjust for as many of the remaining differences as necessary in the longitudinal spouse and control cohorts. The covariates included socioeconomic status, US region of residence, hospitalization during the index month, frailty level for the index month, select pre-index chronic conditions, and length of follow-up. This analysis also was restricted to initially community-dwelling couples. Despite these adjustments to control for pre-index high-cost spouses, a partners’ AD diagnosis still had a substantial independent association with the spouses’ healthcare costs.

In response to Dr. Somme’s comment and to reflect the changes in Table 4, we have revised the first paragraph of the section entitled “AD spouses’ healthcare costs” to read as follows (page 11, lines 201-209):

A sizable cost difference occurred between the cross-sectional AD spouses and matched control cohorts (Table 4). Mean annual Medicare expenditures in the two cohorts were $8,206 (95% CI: $7,967, $8,445) and $6,640 (95% CI: $6,414, $6,866), respectively (P<0.001). The mean total Medicare expenditures are large relative to the medians in both populations, indicating that high-cost outliers – whose records include substantial hospitalization – considerably affect these means.

In the longitudinal cohorts, a regression analysis (Table 4) adjusted for pre-index factors that drive healthcare costs to high levels. Even with these adjustments, AD spousal status was associated with an estimated 29% independent cost impact on Medicare expenditures (P<0.001).
We also revised the sentence on page 9, lines 166-168 to note the regression model’s stepwise construction process:

This analysis, constructed in stepwise fashion, included as covariates socioeconomic status, national region of residence, index year, select pre-index (but not post-index) chronic conditions, annual JFI level and status as AD spouse or control.

3. I have concerns about the use of the “frailty” (and even more “frailty index”, or “score”, the denomination is not always the same) in the paper. The tool used for the purpose of measuring frailty is not in accordance with the current consensus definition of the concept of frailty. It is more a “comorbidity” measurement (with a mobility dimension) than a “frailty” measurement. For example since the cognitive decline is one of the aspects of the “frailty score”, it is not surprising that the AD patient score is different from other groups in table 4 (it is quite circular). Considering that frailty index is currently used in the literature for another tool related to the consensus definition, I would prefer that the authors used a more appropriate denomination (comorbidity index or another expression). To the less, the authors have to add in the discussion that they do not have information on “frailty” of the spouses notably before the AD diagnosis first appearance in claims. It is possibly a explaining factor of the variability of the health care cost in the AD spouse group.

Authors’ response: Dr. Somme critiques the use of the JEN Frailty Index as presented in the old version of Table 4. When inserting the additional cost information to Table 4 mentioned above, we also removed the JEN Frailty Index data. The precise JEN Frailty Index results are extraneous to the present manuscript, although they contribute to the regression analysis at the bottom of Table 4.

We eliminated the columns for the AD patients and the spouses of the controls. Dr. Somme’s comment about the “circular” use of the JEN Frailty Index, although relevant to the AD patients, no longer applies to the revised manuscript that we are submitting.

In addition, the JEN Frailty Index is not simply a comorbidity index. We have edited the manuscript section describing the JFI to make clarify the Index’s nature (pages 8-9, lines 138-159):

We found it necessary to include a covariate indicating each person’s pre-index well-being when constructing regression models of the AD spouse and control cohorts’ healthcare costs. Finding an appropriate marker for patients’ overall health and physical status poses difficulties because of Medicare claims records’ lack of clinical data. In addition, the scope of potential comorbidities in this elderly study population is not adequately represented in a standard comorbidity index such as Charlson or its adaptations [28,29] that cover only a limited number of hospital diagnoses. Another option, stratifying by specific comorbidities, would have been quite challenging statistically. We chose instead to employ the broad-ranging JEN frailty index (JFI). The JFI takes into account claims filed by all providers, not just hospitals. It is therefore
particularly useful in patients with complicated clinical profiles. Furthermore, this index was developed for specific use in Medicare/Medicaid database analyses as part of the Medicare/Medicaid Integration Project, funded by Robert Wood Johnson Foundation’s [30]. We have found it to be significantly correlated with Medicare and Medicaid expenditures as well as mortality [31,32].

The almost 1,800 diagnoses included in the JFI are classified into 13 condition categories. The 13 condition categories are designed to provide an indication of individual patient impairment. They include minor ambulatory limitations, severe ambulatory limitations, cognitive developmental disability, chronic mental illness, dementia, sensory disorders, self-care impairment, syncope, cancer, chronic medical disease, pneumonia, renal disorders and systemic disorders (e.g. septicemia). A patient’s personal index is derived by counting the presence of these condition categories in the previous year’s diagnostic claims.

We appreciate Dr. Somme’s comment that we should not label the JFI a “frailty index.” There are a number of indexes and scores whose name contains the phrase “frailty index,” and we do not want to add to the ambiguity of the phrase. However, we have already used the JFI label in several reports. JFI’s distinction is that it is based on administrative records, not clinical ones. This is an important innovation since claims records from Medicare and other payers continue to be more readily available than clinical records. Claims datasets also contain several advantages over clinical databases. They usually include a much larger population and are more standardized, complete and easily analyzed.

Dr. Somme is not quite correct when he says that we have no knowledge of the pre-index date frailty in the AD spouses. Every AD spouse eligible for the longitudinal cohort was required to have at least 12 months of Medicare claims records available prior to the date of their spouses’ initial AD diagnosis. This information is visible in Figure 1, which shows that mean Medicare expenditures were equal in the AD spouse and matched controls until two months before the index AD diagnosis. The incident spouses and controls’ JFI score on the index AD diagnostic date was a covariate in the regression analysis shown in Table 4. That score takes account of the diagnoses received by the study population in the year prior to the index AD diagnosis.

**Joseph Menzin:**

The authors provide an interesting analysis of the mean costs for spouses of AD patients versus matched controls. The excess cost was reported to be about $150 PPPM higher among the AD spouses. The ability to identify spouses of patients with particular conditions in the Medicare files would be of interest to the journal’s readership.

**General comments:**

(1) Neither the introduction nor the discussion provides any references to other literature that has assessed spousal costs for patients with AD. I would
imagine that others have addressed this issue, so this needs to be carefully discussed and clarified.

*Authors’ response:* As opposed to research on caregivers that typically includes a mix of children, spouses, and other friends/relatives, there are very few studies about the influence of AD on spouses’ health specifically. We already cite the two most relevant studies on pages 4, line 53 (see references 20 and 21). A new, related study is Suehs et al., *J Am Geriatr Soc*, 2014. We have added a mention of this study (page 5, lines 55-56).

(2) One of the main limitations of the study is generalizability of findings to the general elderly or Medicare populations. There seem to be a few major issues, some of which are touched upon but would benefit from further analysis. First, the authors are capturing data from spouses with <10 years of work history. What do we know about this group compared to those with a longer work history—are there any statistics? What might we expect the differences to be? Second, this analysis focuses on the Medicare FFS population. How many Medicare beneficiaries were enrolled in health plans during the study period? How do the two populations compare?

*Authors’ response:* One can only speculate about the differences between previously working and non-working spouses. We discuss the possible differences between the two groups on page 14, lines 272-276.

At the time of this study (2001-2005), membership in Medicare managed care ranged from 13% to 15% of the total Medicare population. There has been a long discussion – and considerable controversy – over the ability of Medicare managed care plans to disproportionately enroll relatively healthy Medicare beneficiaries (see, for example, McGuire et al., *The Millbank Quarterly*, 2011). The relatively small size and healthier status of the managed care population probably limit the effect of its exclusion on our study results.

(3) Most readers will not be familiar with the JEN frailty index. Has it been validated? How does it correlate with the Charlson and Elixhauer comorbidity scores? It would be helpful to provide ICD-9 codes in an appendix.

*Authors’ response:* See our response to Dr. Somme’s comment no. 3 above. It is not practical to include ICD-9 codes behind the JEN Frailty Index because there are about 1,800 of them.

(4) The $150 per month higher costs for AD spouses is statistically significant, but appears modest. Do the authors know what is driving the cost
differences (e.g., hospital, SNF, ER visits)? Also, what are the implications of not knowing drug costs (e.g., AD caregivers may be more likely to use certain drugs, like antidepressants, which would not be captured)

*Author’s response:* The main cost driver for the observed difference was acute inpatient care. The lack of pharmacy data indicates that the estimated difference is conservative. A new study of elderly cohabitants of AD patients (Suehs et al., J Am Geriatr Soc, 2014) was able to include drug costs. These costs turned out to be the same in the cohabitants as in the matching controls. (Hospital costs were not listed separately in Suehs et al., but the cohabitants had significantly higher average total costs than the controls: $7,168 vs. $6,301, respectively).

(5) Even though patients were required to have 6+ months of enrollment in any given year, was there differential mortality for AD spouses and their controls? If so, how might have affected cost estimates?

*Author’s response:* The difference in mortality between AD spouses and controls was not statistically significant (18% vs. 14%, respectively). Mean follow-up time was 11.8 months in both the AD spouses and the controls (See table 4 in the manuscript). We did not separately investigate healthcare costs in the final months of life. Differences in mortality could affect costs in opposing directions. The high costs associated with dying might raise individual monthly costs in the disproportionately affected cohort. Conversely, if high-cost frailer spouses died earlier in the observation period, then long-term average costs might decrease.

Other comments:

(1) Some parts of the manuscript refer to spousal caregivers, spousal health, or spousal caregiver burden. This is questionable since we don’t know if the spouses are caregivers and health status is not reported. Are there data on the % of spouses of AD patients who are primary caregivers?

*Authors’ response:* Thank-you for raising this point. Past studies have found that spouses, when present, contribute heavily to caregiving (see page 4, lines 43-45 and pages 15, lines 295-299). However, our study had no way to directly measure caregiving burden. Many studies conflate spouses with caregivers, but our explicit goal was to measure the spousal experience. Future research will attempt to elicit how much of this effect is due to caregiving burden. We have removed several inadvertent references to our spousal population as “caregivers.”
(2) The terminology of "confirmed AD" should be avoided since there are no clinical assessments available in the claims data.

Authors’ response: We changed “confirmed” to “corroborated” throughout the manuscript.

(3) In Table 1, 86% of AD patients are White. Is this consistent with general elderly population or other AD studies in the Medicare population?