Author’s response to reviews

Title: The Effects of an Integrated Care Intervention for the Frail Elderly on Informal Caregivers: A Quasi-Experimental Study

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Version: 5 Date: 1 April 2014

Author’s response to reviews: see over
Subject: Manuscript revisions

Dear Editor,

I hereby resubmit the manuscript titled ‘The effects of an Integrated Care Intervention for the Frail Elderly on Informal Caregivers: A Quasi-Experimental Study’ by Benjamin Janse, Robbert Huijsman, and Isabelle Natalina Fabbricotti to BMC Geriatrics. One important adjustment to the manuscript is the addition of an author. In the previous version this author was erroneously omitted; this has now been corrected. We have made the requested revisions to the manuscript and, where needed, provided additional explanation in response to the reviewers’ questions/comments.

Reviewer R. Melis

We want to express our grateful to this reviewer for the substantial inputs of time and energy that have obviously been spent reviewing our manuscript. His comments have truly helped us to improve our manuscript.

Major compulsory revisions:

1. In the introduction section, more emphasis is given on the possibility of adverse effects of integrated care on informal caregivers (see lines 77-91). Also, the hypotheses and research question are now formulated accordingly (see lines 106-111).

2. We did not use a hierarchical structure or division of the outcomes into primary and secondary outcomes. The reviewer is absolutely right that this needs to be mentioned explicitly in the manuscript, and this is now added to the main text (see line 188). Within and between-group analyses were performed to provide insight into scores on outcomes and must be considered as descriptive data. No treatment effect can be derived from these data, and thus we performed regression analyses to enable to determine the effect of the WICM controlled for the control variables. This has been clarified in the manuscript (see lines 253-261 and lines 263-275).

3. See the answer in comment 2.
Minor Essential Revisions:

4. Reference to paper regarding the development of the questionnaire has been added.

5. This variable was binary (yes/no) and thus, linear regressions were no option. Instead logistic regressions were used. This point is now emphasized in the methods section (see lines 265-269).

6. Models and effects of the WICM were considered significant if $p<0.05$. However, it is not uncommon to allow apply less rigid significance levels when sample sizes turn out to be relatively small. This rationale is now added to the manuscript, including a reference (Cohen, 1988) (see lines 276-277).

7. There was some concern of multicollinearity as there was a risk of overlap between independent variables, e.g. education and income are both indicators of SES. Thus, in each regression that was performed, the corresponding measures were checked for the model containing the intervention (Model 3). For linear regressions, these measures are tolerance and VIF, for logistic regression this is the standard error. In the manuscript, explanation regarding multicollinearity is now extended and contains a reference (Fields, 2009) (see lines 778-283).

8. The reviewer makes a strong point by emphasizing the importance of taking practice effects into account. We actually considered including ‘practice’ as a control variable in the regression analyses. We thus performed exploratory regression analyses including practice as control variable, but this added no value to the strength of the regression models. Moreover, practices differed greatly in size and therefore also the participating patients/caregivers they provided, ranging from 10 patients to more than a 100 patients and thus yielding extremely skewed distributions. Additionally, we were primarily interested in the differences between experimental and control practices. Ultimately, it was decided against including ‘practice’ as a control variable.

9. -In the recruitment process, a basic definition was indeed used to identify informal caregivers. The participating patients were asked if they received non-professional and unpaid care from partners, family or friends. While identification thus occurred based on this definition, some informal caregivers did not see themselves as such. For instance, when tasks were transferred to formal caregivers when the patient moved into a nursing home. This disqualified them as participants in this study. In the ‘participant’ paragraph of the methods section, this is now described (see lines 156-167).

- The reviewer makes a valid point. The reason was that unwilling informal caregivers were mostly the partner of a patient that withdrew from the study prior to T1. Most of
these caregivers indicated that if their care receiver did not want to participate, they didn’t want to participate either. To enhance the understandability of this section we have reformulated this section and aggregated the losses to follow-up due to ‘unwillingness to continue’ into one category (see lines 296-302).

10. –It is now described more clearly that the definitive groups were determined after losses to follow up; only those respondents with data for both T0 and T1 were included into the final sample. Groups from the final sample were compared in terms of control and outcome variables at T0.

-The reviewer puts forth a very important issue, and we acknowledge the need for an analysis of non-responders. However, around 50% of non-responders dropped out before a baseline measurement was performed, which means that the data needed to assess the effect of the loss to follow-up are not available. This point is now described in the discussion of the manuscript (see lines 510-516).

11. Answer: of the caregiver, this is added to the text in the manuscript (see lines 312).

12. Table 1 now contains background data of the care recipients and the manuscript now includes a description of these data and differences between groups. Data regarding background characteristic of the informal caregivers that was previously part of Table 2, is now included in Table 1. Also, it now contains information about the relationship between caregiver and care recipient. Table 2 also contained T0 scores; however, as Table 3 basically provides the same information, Table 2 has been eliminated from the manuscript.

13. The steps from going from 184/193 to 144/118 are not related to non-participation, but indicate how much of the care recipients received care from an informal caregiver. To clarify this point, this is now more underlined in the description in the manuscript (see line 294).

14. The study population was generally similar to the general population of Dutch informal caregivers on all background characteristics (SCP report, 2010 [in Dutch]). Therefore, we do not feel that generalizability is affected.

15. -It is not uncommon that within-group variations exist without resulting in any between-group differences, as between-group analysis is about comparing difference scores. While this may sometimes yield contradictory findings, independent T-tests provide a solid measure for between-group comparison.

-As suggested, a footnote has been added for the “–“ in table 2.

-We understand the reviewer’s suggestion of using an ANCOVA and agree that an ANCOVA would have done the job as well. However, regression analysis and ANCOVA share the same statistical model (linear regression model); thus, to achieve the aims of the current study both methods are exchangeable. Our regression models
were similar to what is suggested by the reviewer i.e. an outcome Y with baseline scores, control variables and "yes/no intervention" variable as predictors (see e.g., Fields, 2009).

Table 3 shows the results of the regression analyses after controlling for baseline scores and control variables. As a Final step (model 3), the intervention (yes/no) variable was added and thus, the effect of the intervention is shown. To clarify this, the Table’s has been formulated to accordingly.

For comprehensiveness and clarity, Table 3 now contains all coefficients/ Exp (B) values of all predictors (including non-significant effects) and significant coefficients are shown in bold.

16. Table 3 now shows the range for each outcome, a more informative title and a note explaining the interpretation of the results shown (Please note that the Table is according to the author guidelines of the journal).

17. The reviewer puts forth a very interesting issue. We share his interest in effects on different subgroups of informal caregivers, especially regarding co-residence. However, as subgroup and interaction effects were not hypothesized a priori, it would not have been appropriate to report them in this study. In addition, due to the relatively small sample and large number of predictors, any effects resulting from subgroup and interaction analysis would have been subject to serious methodological limitations, such as capitalization of chance (Lagalos, 2006).

18. Each consecutive model of the regression analyses i.e. all regression analyses were designed using 3 models (as explained in the methods section): Model 1 containing the baseline scores, model 2 containing the control variables and model 3 containing the intervention. In the regression analyses, the effect of the intervention was assessed by investigating the output from Model 3. As suggested by reviewer ms. Janevic, table 5 is now converted into Additional File 2, its contents now being briefly discussed in the regression paragraph of the results section (see lines 394-397).

19. As part of the evaluation of the WICM, several process indicators have been assessed to determine the degree of effective integration resulting from the intervention. However, the results of that process evaluation deserve more space than would be allowed if they were reported in the current paper. Therefore, the process evaluation will be reported in a separate paper to be published in the near future. However, we agree with the reviewer that the degree to which the intervention was actually
delivered, provides a possible explanation of the relatively limited effects of the current study and is a fundamental discussion point. Therefore, in the discussion additional text has been added that briefly touches upon this point (see lines 476-480).

20. We agree with the reviewer that incorporating the care recipient's status into the analyses would make our results stronger. However, we explicitly aimed to include only caregiver characteristics as we considered the amount and of variables that could be of influence too high. As the reviewer indicated earlier, practice effects would also qualify, as did the frailty scores of the patients.

Reviewer Joanie Sims-Gould
We thank the reviewer for her valuable comments. Especially her suggestion to reduce the number of limitations has helped us to improve the clarity of the discussion substantially.

Major Compulsory Revisions:
- As suggested, we have reduced the list of limitations to 4 main limitations. In addition, the point of co-residency is added to the discussion of results as a potential explanation (see lines 468-474) and this point has been more emphasized in the recommendations (see lines 533-535).
- The use of the word 'indeed' has been reduced in the manuscript

Reviewer Mary Janevic
We would like to express our appreciation to the reviewer for her many valuable comments and the considerable amount of work that it must have demanded. We believe that her comments, questions and insights have improved this manuscript substantially.

Discretionary Revisions:
1. In the manuscript, a definition of frailty has been added (see lines 50-53).
2. In the description of the intervention, additional information is provided regarding the available services for informal caregivers, and what the WICM offers to informal caregivers in terms of assistance and types of support (see lines 136-143).
3. Yes, a study of patient outcomes was indeed part of the evaluation. These results will be published in the near future.
4. Table 1 now contains information of both care receiver and informal caregiver.
5. We agree with the reviewer that the informational value of Table 5 is limited. Therefore, this table is converted into Additional File 2. Its content is briefly discussed in the regression paragraph of the result section (lines 394-397).

Compulsory Revisions:

1. Perceived health has been measured on a 5-point Likert scale. Consequently, these data were transformed into a 100-points scale as prescribed by the guidelines of the RAND-36, from which these items were obtained (Dutch Version: Van der Zee, 2012). To clarify this transformation and transformations performed on other outcomes, a paragraph has been added to the analysis section (see lines 239-250).

2. All caregivers (control and experimental) were recruited in the same way i.e., all patients of 75+ of the participating practices were approached. Informal caregivers could only be included if they provided care to a patient that participated in the intervention.

Additional information has been added to the methods section, describing more clearly the recruitment of patients and caregivers, including the recruitment of control caregivers (see lines 156-167). In addition, Response rates and characteristics of the participating patients have been added in the results section (see lines 287-293).

3. The background section expanded with a discussion of possible effects of integrated care (both positive and negative) for all outcome measures including health and time spent (see lines 77-91). However, due to the scarcity and consistency of existing literature, we considered it less appropriate to formulate hypothesis regarding changes in the nature of tasks.

4. Yes, we have gathered data regarding several patient outcomes, including health status. However, in the current study we limited the control variables to caregiver-related characteristics. The patient outcomes will be the subject of future papers. This is now described briefly in the discussion of the manuscript (lines 502-508).

As suggested by the reviewer, Table 1 now also shows the frailty scores of participating patients.

The relationship to the care recipient was in fact considered to be an important control variable and these data were gathered (child/spouse). However, multicollinearity analyses revealed that this variable showed substantial overlap with the variable ‘living with care recipient’, which indicated that their predictive value was largely exchangeable. It was decided to drop ‘relationship’ as a control variable. This has now been described in the manuscript (see lines 278-283).
5. We agree with the reviewer that the within- and between-group analyses do not contribute to answering the research question. These analyses are not meant to do so; they are reported solely for descriptive purposes. While they show the developments in scores for both groups, our primary goal was to assess the effect of the intervention using regression analyses. However, the reviewer makes a valid point regarding the notable between-group patterns. While most of these patterns are not the result of the intervention (as shown in the regression analyses), the question can be raised whether this remains the case over a longer period of time, or whether some other variable is responsible. This is now briefly discussed in the discussion of the manuscript (see lines 478-489).

6. For both items regarding perceived health, we followed the guidelines of the RAND-36 instrument, from which these items were derived. The guidelines prescribe how to transform the original 5-point scale into a 100-point scale using a particular formula. The reviewer is referred to these guidelines for further reading (Dutch Version: Van der Zee, 2012; see also Hays et al., 1993). A paragraph describing these transformations has been added to the analysis section (see lines 239-250).

7. The reviewer rightly underlines that caution is warranted in interpreting data based on a quasi-experimental design. However, by performing multiple regression analyses the main limitations of such a design are countered. Moreover, regression models allow inference of causational direction to a certain degree (Fields, 2009).

8. As the CarerQoL describes the caregiver situation by assessing the problems/benefits resulting from care provision and is described by developers of the instrument as a burden instrument (Brouwer et al., 2006), it must be considered a measure of subjective burden. Conversely, we included separate items to assess the overall quality of life.
   -Regarding household tasks: It concerns whether an informal caregiver performs these tasks (yes/no). We agree that in the table showing the results of the regression analyses, the names are confusing. Therefore, we changed the names to ‘caregivers performing household/personal care/instrumental care tasks’

9. The manuscript has been checked for consistency in outcome names; organization and categorization of the Tables have been made identical.

10. In the discussion of the effect observed on subjective burden we have added a more detailed description of what this effect entails. With this addition, we attempted to make the distinction between CarerQoL and quality of life more clear (see lines 414-417).

11. This regards yes/no data at 2 points in time. In the within and between table, the percentages of caregivers indicating ‘yes’ are shown i.e. those who indicated to perform household, personal care and instrumental care tasks. The binary character of these
data required logistic regression analyses rather than linear regression analyses. Outcomes thus must be interpreted in terms of likelihood.

- While it was hypothesized that tasks would change, hypothesis concerning the nature of these changes was less straightforward as literature on the subject is not consistent.
- We agree with the reviewer that the statement regarding ‘improvements in care receivers status’ and personal care tasks was not warranted. This phrase is now formulated in more general terms (see lines 449).

12. The reviewer puts forth a very important issue and therefore we have now extended the discussion regarding loss to follow-up (see lines 510-516).

13. The suggested limitation has been added in the manuscript.

14. As subgroup and interaction effects were not hypothesized a priori, it would not have been appropriate to report them in this study. In addition, due to the relatively small sample and large number of predictors, any effects resulting from subgroup and interaction analysis would have been subject to serious methodological limitations, such as capitalization of chance (Lagalos, 2006). Nonetheless, the reviewer puts forth a very interesting point which we will definitely consider in future research protocols.

15. In the discussion, some issues of preserving the control group are added (lines 489-491).
References:


http://www.scp.nl/Publicaties/Alle_publicaties/Publicaties_2010/Mantelzorg_uit_de_doeken