Author's response to reviews

**Title:** How are patients managing with the costs of care for chronic kidney disease in Australia? A cross-sectional study

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**Author's response to reviews:** see over
2 November 2012

Dr Hayley Henderson
Executive Editor
BMC Nephrology

Dear Dr Henderson,

Original Manuscript ID MS: 2051439938739133

Manuscript TITLE: How are patients managing with the costs of care for chronic kidney disease in Australia?

Thank you for your email dated October 29th 2012 and for the opportunity to respond to the reviewer’s comments about the above mentioned manuscript. The reviewer’s comments were very useful and have helped to improve this paper. We have outlined the changes made to the manuscript below. We look forward to the opportunity to have this manuscript published in BMC Nephrology.

Please contact me on either +61 2 9993 4562 or beverley.essue@sydney.edu.au if you require additional information.

Sincerely,

Beverley Essue

Co-authors: Germaine Wong, Jeremy Chapman, Qiang Li and Stephen Jan
Reviewers’ comments and our response to each comment are summarised below:

Editorial requests:

a. Please remove the "Main outcome measures" heading from the abstract.

b. Please change the abstract section heading "Background and Objectives" to "Background"

c. The objectives statement may be added to the abstract Background section.

a-c requests have been completed.

Reviewer 1: Jennifer Zelmer

Major Compulsory Revisions

a. The overlap between the two components of the sample (patients invited to participate by mail and those invited to participate from community dialysis centres) is not clear as the methods section is currently written. It is specified only for those who were invited to participate from community dialysis centres but had already received and completed the study questionnaire by post. As a result, the overall participation rate calculation is not clear. Equally, it is not clear what steps were taken to ensure that there were no duplicate responses between the two groups.

This has been clarified in the methods section with the addition of the following sentence in the second paragraph of the Methods section:

“Individuals who had already completed the questionnaire by post were not invited to participate again.”

b. The findings described in the paragraph before the discussion section, i.e. that out-of-pocket spending was not associated with economic hardship as measured in this paper, are possibly counter-intuitive but significant in the context of the topic being discussed. They deserve to be mentioned in the abstract as they fundamentally affect the interpretation of results. Consideration should also be given to further analysis of these findings in the discussion section.

Thank you. To clarify, the relevance of this issue is discussed in the second paragraph of the discussion section. In addition, the following sentence has been added to the last sentence of the results section of the abstract:

“Out-of-pocket costs and financial catastrophe were not found to be significantly associated with hardship in this analysis.”

c. Reference 19 is used to support a definition of a catastrophic level of spending as an out-of-pocket burden greater than 10 percent of equivalent income (page 7) but that study used a different definition (household financial contributions to the health system greater than 40% of income remaining after subsistence needs are met).
We thank Reviewer 1 for this correction. The reference has been replaced with the following reference:


Minor Essential Revisions

a. The abstract would be clearer if the definition of catastrophic spending levels used in this paper (i.e. out-of-pocket spending greater than 10 percent of equivalent income) was included.

As suggested, we have included this definition in the abstract.

b. The abstract indicates that various factors “determined” hardship. As the nature of the analysis performed does not permit causal inferences, it would be better to use a different term.

We thank Reviewer 1 for this correction. The term “associated” has been used instead throughout the manuscript.

c. The article is generally well-written, but there are some remaining typographical issues (e.g. “an” instead of “and” in the first sentence of the introduction and “to” rather than “and” in the methods section of the abstract). Although copy edit would be helpful to ensure that any remaining issues are addressed before the article is published.

Thank you. All typographical errors had been corrected.

d. The format and legends of some figures could be strengthened to improve readability, e.g. figure 2 and 4. Likewise, as readers will likely view/print figures in black and white, shading/presentation decisions should be made accordingly to ensure clarity (e.g. figures 3-6).

The shading in Figure 2 and 3 have been revised to improve readability when printed in black and white. Based on Reviewer 3’s comments, the data presented in Figure 4 have been included in a table instead of figures. Figure 4 has been removed from the manuscript.

e. Reference 4’s analysis is based on a cross-sectional design. It is not clear why this reference is used to support the assertion of increases in health expenditure for chronic illness.

This sentence has been revised to clarify the point being raised. The reference has also been changed:

“Against this is a background of increasing health expenditure in the form of out-of-pocket payments [4].”

AIHW, 2012

f. Reference 5 cites a draft report. It would be preferable to cite the final report, rather than the draft, if available.

The final report for this work is not available in the public domain (as confirmed by Healthcare Management Advisors Pty Ltd, the consultants who generated this report). The draft report is endorsed by Kidney Health Australia, the national peak body for the improvement of kidney health outcomes. Due to the unavailability of the final report, this draft report is commonly cited for estimates of out-of-pocket costs for CKD in Australia.

Discretionary Revisions

a. It may be helpful to put the findings from this study into context with cost-related access issues reported by Australians, including those with chronic illness, more generally. For example, surveys from the Commonwealth Fund include this type of information (see http://www.commonwealthfund.org/Topics/International-Health-policy/Bar.aspx?ind=503&loc=53 for instance).

As suggested, the following sentence has been added to the first paragraph of the discussion.

“In addition, the burden of out-of-pocket costs found in this study, a mean of AUD$907 in the previous three months, reinforces wider concerns about the extent to which the Australian health care system relies on individual contributions to fund health care. Individual spending on health care in Australia is high by international, high-income country standards. In a recent Commonwealth Fund survey of 11 high-income countries, the incidence of out-of-pocket costs exceeding US$1000 in the previous year among individual respondents was 21% in Australia – behind only the United States (35%) and Switzerland (25%)”


b. The authors suggest that to mitigate the financial hardship experienced by individuals with CKD, clinicians should identify those most at risk of hardship and link them with early support. While this may indeed be a useful intervention, the basis for proposing this intervention instead of other possible options is not clear in the text.

The current study raises the issue of the affordability of care – an issue which should be of concern to clinicians, even if the solutions to improve the affordability of care are outside of their direct remit. Given the potential links between economic hardship, out-of-pocket costs and adherence to recommended care, this suggestion stems from other research (cited in the 2nd paragraph of the discussion section) on the reluctance of patients to raise cost issues with clinicians as well as clinicians’ lack of awareness of cost as a barrier to adherence. We suggest this option as one tangible way forward for improving support for patients who are at risk of hardship to access the systems of support that may already be in place.
c. The authors may wish to consider whether the format of response (written questionnaire versus assisted completion in the dialysis centre) has any effect on results. Equally, the authors may wish to consider whether there are any gender-related differences related to the analysis in Table 3.

Thank you for this comment. I refer the reviewers to the methods section of the paper that describes the modelling strategy used in this analysis. Variables for ‘mode of survey completion’ (self-completion versus completion in the dialysis centre) and gender were included in the initial model as potential explanatory variables. However, neither of these variables was significant for inclusion in the final model.

d. In the last sentence of the first paragraph of the introduction, is there evidence to suggest that these expenses are unbudgeted? This is undoubtedly true for some patients but may not be for others.

As suggested, the term ‘unbudgeted’ has been removed.

e. The results in figures 4-9 may be clearer and more concise if presented in tabular form.

This change has been made. The results now appear in Table 3.

f. As currency exchange rates have been fairly variable in recent years, it may be helpful to include a timeframe for the notional exchange rate shown, e.g. in figure 4.

The following has been added to the footnotes in Table 1 and 3 and to the first paragraph under the heading “Relationship between out-of-pocket costs and hardship” in the results section.

“AU$1=US$1=EU€0.73=GBP£0.62; January 2011”

g. Consider including the questionnaire used as online supplementary material

As suggested, a supplementary table has been added (to be available online only) that describes the domains of the study questionnaire.

Reviewer 2: Louisa Gordon

Major compulsory

a. Labelling the study ‘prospective’ is true in the sense of participant recruitment but the data itself is captured retrospectively so I am not sure it is entirely accurate to say it is a prospective study. The descriptor ‘cross sectional’ is perhaps better.

As suggested, we have replaced the word “prospective” with “cross-sectional” in the title and first line of the methods section in the abstract.

b. Abstract – the p values are not necessary when the 95% CIs are presented – usually present one or the other.
The p-values have been removed.

c. **Abstract** – the results could be clearer because currently all factors influencing hardship are lumped together. It would be clearer if the protective factors were separated from the negative factors.

Agree, this revision has been made to the results section of the abstract.

d. There are some typos in the manuscript that should be corrected. E.g. introduction, 2nd line, p9 starting a sentence with ‘19%’ is incorrect - should be in full text.

Thank you for this comment. The paper has been edited and all typographical errors have been corrected.

e. Can the authors provide a rationale for the recall period of ‘previous quarter’? Does this adequately capture most costs that would accrue? This time period needs to be included in the graph titles.

There is a lack of consensus on the appropriate recall period that should be used in surveys to collect expenditure and health utilisation data and as a result, a variety of disparate approaches are used across most studies [1-3]. A review of the literature highlights two key trade-offs: longer recall periods (i.e. 12 months) tend to lead to measurement error and underestimation of expenditure (due to forgetting), while shorter recall periods (i.e. 1-4 weeks) tend to overestimate expenditure (due to telescoping) and may not be a true reflection of usual consumption [1-4]. The debate in the literature in this area also indicates that no single recall period will be optimal for collecting all types of data on health spending [5] and there is an inherent trade-off between accuracy and information loss when selecting the recall and reference periods that will be appropriate for a given study [1]. Reviews of this literature indicate that in the absence of validation studies, researchers should select reference and recall periods that are appropriate for the type of expenditure data collected in the survey.

This study adopted a reference and recall period of three months (previous quarter) for collecting data on out-of-pocket costs. This time period was selected given the chronic and progressive nature of chronic kidney disease (CKD) and the routine pattern of medication and health service utilisation required for the effective management of CKD. In order to avoid over-estimating true expenditure, all cost data are reported for the stated period (three months) and not extrapolated to 12 months.

This reference period will be added to the title of Table 3 which replaces Figure 4 (this replacement was made based on the advice on reviewer 1).

We appreciate the reviewer’s suggestion. However, on review of the Briggs and Gray 1998 reference cited, we believe that the Mann-Whitney U test is appropriate in this analysis given that the main focus of our analysis is on hypothesis testing, rather than estimation. In addition, based on the advice of Reviewer 1 and 3, we have included both means (SE) and medians (IQR) for the cost data and include the test statistic from both the parametric and non-parametric hypothesis tests. These results are now included in Table 3.

g. Analyses – I think it would be interesting if another sub-group was those participants with comorbidities (e.g. <3 vs >=3). This could shed light on the impact of concurrent conditions and these costs for patients particularly diabetes, known to have high out-of-pocket costs.

Thank you for this comment. I refer the reviewers to the methods section of the paper that describes the modelling strategy used in this analysis. A dichotomous variable for co-morbidity (yes/no) and continuous variable for number of comorbidities were included in the initial model as potential explanatory variables. However, neither of these variables was significant for inclusion in the final model.

h. Discussion – page 12, top paragraph lists the results of the financial hardship questions for the first time. I think this belongs in the results section.

To clarify, the results for the economic hardship outcome are reported in the results section (under the heading: “Hardship and CKD”). The first paragraph of the Discussion section compares the results of the current study with other findings in Australia (in chronic disease populations and the general population) and international studies in order to put the results of this study into the context of the literature in this area.

i. Discussion – the authors state on p13 that a prospective design with measurements taken over time will inform change in financial circumstances etc. But the patients are already, on average, 5 years past their dialysis and 12 years past their diagnosis – so these costs may already reflect the situation of the established ‘steady state’ or perhaps best case – those not working by now may never do so? The authors may wish to comment along these lines.

As suggested, we have added this important point as “limitations”. (page 13, last paragraph of the discussion section). The addition reads:

“Given that our study population had been receiving dialysis for a mean of five years and were on average 12 years past their diagnosis, our results likely reflect the situation of the established ‘steady state’ for the households affected, demonstrating a need for early interventions to assist households to better cope with the negative economic sequelea of CKD.”

Minor essential

a. Abstract – the p values are not necessary when the 95% CIs are presented – usually present one or the other.

These have been removed.
b. There are some typos in the manuscript that should be corrected. E.g. introduction, 2nd line, p9 starting a sentence with ‘19%’ is incorrect - should be in full text.

These have been corrected.

c. Can the authors provide a rationale for the recall period of ‘previous quarter’? Does this adequately capture most costs that would accrue? This time period needs to be included in the graph titles.

The justification for this recall period is discussed above. The time period has been added to all tables.

Reviewer 3: Laura Plantinga

a. The authors’ third aim in the Introduction (examining outcomes by hardship) is never presented in the paper (rather, QOL is used as a predictor of hardship). I would suggest deleting this aim, particularly since the authors present it as a descriptive study.

As suggested, we have removed this statement from the “Introduction”.

b. Self-report of out-of-pocket expenses: it is probably difficult to remember exact numbers for some of these expenditures, particularly if the questionnaire is filled out at the clinic without access to bills and records. Do the authors think they are under- or over-reported?

Self-report is a commonly used mode for collecting cost data, particularly out-of-pocket costs, in economic analyses [2,4,6]. The authors acknowledge that there can be challenges and bias associated with the recall of exact expenditure data and this is a common issue that is hotly debated in the economic literature. We have addressed our approach to minimise this bias in our response to Reviewer 2’s major point (e) above.

c. As the authors point out, the participation rate is somewhat low, at 63%. Non-response is likely related to both outcomes and to hardship, giving a potential for bias in associations. This should be stated explicitly in the limitations, as this is a separate issue from generalizability.

The following sentence has been added to the limitations (second last paragraph of the discussion section):

“The observed non-response is likely related to both outcomes and to hardship status, giving a potential for bias in association.”

d. Which variables were tested for effect modification? Were there a priori hypothesis about interactions? What were the results (presumably null since no interaction terms are presented in the tables)?

To clarify, effect modification was checked between all variables in the model to identify interactions that were significant at the level of $P<0.01$. None of the interactions were
significant for inclusion in the final model. The following statement can be found in the methods section of the paper:

“Effect modification was checked between variables in the model to identify interactions that were significant at the level of \( P<0.01 \).”

**Minor Essential Revisions**

**a.** In the abstract it is stated that variables at the community level are examined—-I didn’t see this in the paper. Should this statement be removed?

This has been corrected. The last sentence of the methods section in the abstract now reads:

“Multivariate analyses assessed the relationships between economic hardship and individual, household and health system characteristics”

**b.** The authors state that the questionnaire and opt-in was mailed to a “cross-section” of patients in Western Sydney. More detail is needed. How were the patients identified? Was it a random selection? How many clinics/locations were involved?

To clarify, convenience sampling was used to identify the cross-section of participants for the mailed survey. As mentioned in the methods section of the paper, individuals were current recipients of care for CKD III-V in Western Sydney, Australia and were across the spectrum of illness, including those receiving renal replacement therapy. At the time that this study was conducted there was a central hub of renal services that operated from one large teaching hospital, Westmead Hospital. This hub of services provided care for all individuals in the Western Sydney catchment (formally, the Sydney West Area Health Service).

The following highlighted statement has been added to the methods section (Methods section, Subjects and Methods sub-section):

“An opt-in invitation and a study questionnaire were mailed to a cross-section of individuals, identified by convenience sampling by clinic staff, across the spectrum of illness including those receiving renal replacement therapy”

**c.** Out-of-pocket costs do not appear to be normally distributed. I would suggest reporting only medians/IQRs and performing non-parametric tests to compare by groups. (Although the authors state that they present means for ease of interpretation, I think medians/IQRs are fairly well-understood).

Medians are now reported for all cost data in Table 3.

**d.** Table 1 is hard to read. Please add % signs where appropriate to distinguish numbers from percentages.

Percentage signs have been added.
e. *Table 2 is probably unnecessary since it doesn’t address the study question. The potential lack of generalizability can be described in the text.*

Table 2 has been moved to an online only supplementary results section.

f. *Figure 4: what do the bars represent? Again, I think medians/IQRs might be better here. These numbers may be easier to interpret in a table since it is difficult to distinguish the hardship/no hardship groups.*

The data in figure 4 now appear in Table 3.

**Discretionary Revisions**

a. *Although a conversion factor to $USD is listed in the table legend, it might be helpful to the international reader if some of the costs are presented both in Australian and other currencies (e.g., U.S. dollar, British pound, euro) so that the relative amounts are better understood. Or perhaps they could be presented as relative amounts as well (e.g., % of income).*

The following currency exchange rates have been added in the text and as footnote to Table 1 and 3:

“AU$1=US$1=EU€0.73=GBP£0.62; January 2011”

b. *Hardship was a dichotomous outcome. Was there any way to calculate a degree of hardship from the various “dissaving” behaviours? It might be interesting to explore whether there is a dose-response type of association.*

Economic hardship was created as a composite measure to incorporate any instance of financial stress or dissaving behaviour. This composite measure provides a practical approach to measuring the household economic burden of illness. The indicators were not ranked or weighted as households often prioritise expenses and make substitutions based on their individual circumstances. Future work will investigate the potential prioritization of strategies used by households and will provide an opportunity to further explore whether there is in fact a dose response relationship.

**References**


