Author’s response to reviews

Title: When a parent dies - a systematic review of the effects of support programs for parentally bereaved children and their caregivers

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Author’s response to reviews:

When a parent dies – a systematic review of the effects of support programs for parentally bereaved children and their caregivers

Below you will find our responses to the reviewers.

David Trickey (Reviewer 1):

Reviewer comment:

This is a really useful review, which has been well-conducted and well-written. The authors should be congratulated on condensing such a disparate body of research in to such an accessible article.

I do have a few suggestions that in my humble opinion will improve it further.

Authors responses:

Thanks a lot for your valuable comments.
Reviewer comment:

Page 4, line 19: The Webb reference is not the best citation to support the assertion that a closer relationship means a higher impact for a child. The Webb reference states this and then cites three other references to support the statement. It would be better to cite a reference that actually provides support for the assertion rather than one that states it and cites other sources. For example Worden (1996).

Authors responses:

The reference is changed to Worden (1996).

Reviewer comment:

Page 4, line 57: I find the wording of the 2nd inclusion criteria confusing - it could be taken to mean that studies HAD to include children whose parents were cared for in the terminal care phase. I would suggest that it would be clearer if the fact that some studies happened to include such children was listed in the results, or a separate note that this was NOT used as an exclusion criteria.

Authors responses:

We have removed the sentence from the inclusion criteria and added the following sentence in the Results section, Study population:

“The population included children whose parents were cared for in the terminal care phase prior to the parent’s death, as this was not an exclusion criterion.”

Reviewer comment:

Page 5, line 20: In the interests of transparency replicability, this exclusion criteria should include the definition of "so small", together with some justification.
Authors responses:

We have added a sentence that we excluded

“Studies with a population of less than 30 participants”.

Reviewer comment:

Page 5, line 40: I'm not sure that the "common language effect size" is actually very common, and so a brief description of what it is would be useful here

Authors responses:

We have added (p 5) Data analysis:

The CL is also known as the probability of superiority, and represents the probability in percent that a randomly selected person will score a different observed measurement post- than pre intervention, after controlling for individual differences.

It is also explained in the Note of table 2:

“The CL effect size indicates that after controlling for individual differences, the likelihood in percent that a person scores a different observed measurement for Mean 1 than for Mean 2”

Reviewer comment:

Page 8, line 4: I cannot understand "one study focused on expected respectively sudden/violent death" It might just need some punctuation.

Authors responses:

We have tried to make this sentence clearer:

“One study focused on children who had lost a parent to expected versus unexpected deaths”. 
Reviewer comment:

Page 9, line 48 - page 10 line 4: I am not convinced that a simple list of mediating factors associated with just one intervention is very helpful, unless you make the case that these are likely to be generalisable and then discuss practical implications

Authors responses:

We have chosen to remove the paragraph about mediating factors.

Reviewer comment:

Page 10, line 41 (and page 12, line 13): The rather prescriptive conclusion that "interventions need to be directed to both the bereaved child and the child's remaining parent or caregiver", does not follow from the results - unless I am missing something. For example, all three of the interventions that were mainly directed to children (i.e. NOT directed to both) achieved statistically significant results (with the exception of one measure from one study), including one (writing for recovery) which achieved the largest between groups effect-size of any of the studies in the review. In any case, there may be very good reasons why some interventions can NOT be directed to parents.

Authors responses:

We have changed the conclusion to the following:

“One conclusion from this review of interventions is that there are studies that have shown effects for children and their caregivers. The results indicate that supportive interventions can be directed exclusively to the children or to both the bereaved child and the child’s remaining parent or caregiver. Support for the children’s caregivers can strengthen their own health and their capacity to support their children.”

We have also added a sentence about parental support as a protective resource for the bereaved children:
“A supportive parenting is a protective resource for parentally bereaved children” (with a reference to Haine et al. 2008)

Reviewer comment:

Page 10, line 53: The statement that "sessions involving both child children and the remaining parent are an important component" is not really a conclusion from your results. You may say that such session are considered important, or "may" be important. But to state it so starkly really requires some data to support it.

Authors responses:

We have changed this sentence to the following:

“Simultaneous family sessions involving both children and the remaining parent may be an important component in a support program as such sessions are sometimes the first occasion that the parent and children have had the opportunity to sit down together and talk about the loss and their feelings about it”

Reviewer comment:

Page 13, line 7: The PERI is listed out of its alphabetical order

Authors responses:

Thanks for your attention.

The PERI is now listed in alphabetic order.
Reviewer comment:

Table 2: The final column "pow" is not explained or described anywhere

Table 2: Writing for recovery dz is in as 1,26 - it should be 1.26 (presumably)

Table 2: It would make this important table a lot more easy to navigate if the column titles were repeated at the top of each page.

Authors responses:

The column power stands for statistical power. However, after the comment from reviewer 3, we have omitted the statistical power column (see comments to reviewer 3).

We have changed this typing error: 1.26

We will add column titles at the top of each page when we know how the table 2 will be lay outed in the journal.

Alison Penny (Reviewer 2)

Reviewer comment:

Thank you for this important article, which adds to our understanding of the effectiveness of supportive interventions for parentally bereaved children, and makes useful suggestions for further research in this area.

Authors responses:

Thanks a lot for your valuable comments.

Reviewer comment:

P3 Line 9-10 While anxiety and depression are mental health problems, I would not describe perception of lack of control as strongly as this. Perhaps 'risks of mental health problems and threats to emotional well-being...' or something similar.
Authors responses:

We have changed the sentence to the following:

“Several studies have shown an increased risk of mental health problems and threats to emotional well-being for affected children, such as anxiety, depression and a perceived lack of control over what happens in one’s life”

Reviewer comment:

p3 line 18 Brent et al's sample was limited to those bereaved suddenly - this should be specified

Authors responses:

We have added that the youths in the study of Brent et al. (2012) were bereaved suddenly.

Reviewer comment:

p3 Line 35 replace 'lives' with 'live'

Authors responses:

The sentence is changed:

“After the death of a parent some children live with their remaining parent”

Reviewer comment:

p3 42-43 Unhelpful to conflate traumatic and complicated grief here - two different concepts. This paragraph is about child traumatic grief - another may be needed on complicated/prolonged grief in young people (eg Spuij et al 2012).

Authors responses:

We have included a paragraph about prolonged grief with reference to Spuij et al. (2012):
“After the loss of a parent children can also develop a syndrome referred to as prolonged grief disorder that includes a persistent and disruptive yearning. The child has difficulties to accept the parent’s death and difficulties to move on. The syndrome also includes detachment, bitterness, and a sense that life is meaningless.”

Reviewer comment:

p3 line59 If the policy impetus for this paper was specifically about sudden death, does it matter that the studies include those for children bereaved through expected death?

Authors responses:

Even if the Swedish policy gives attention to children bereaved unexpectedly we have chosen to include expected deaths in the review. There are few effect-studies in the field; therefore we do not consider it is possible to conduct a systematic review exclusively about effects from interventions for suddenly bereaved children.

We also believe it is important to gain knowledge about interventions for children bereaved through expected deaths, as these children may need support as well.

Reviewer comment:

p5 line 3-4 Perhaps alter 'whether the intervention had been effective for the children' to 'the effectiveness of the intervention for the children' as this is a matter of degree rather than yes/no

Authors responses:

We have changed the sentence:

“The publications offered any quantitative outcome measures that demonstrated the effectiveness of the intervention for the children”.
Reviewer comment:

p5 line 21 Would be helpful to know if this was based on a particular cut-off/ power calculation, particularly given that this point is used in the discussion

Authors responses:

We have added a sentence that we excluded

“Studies with a population of less than 30 participants”.

Reviewer comment:

p6 line 38 In the phrase 'there was strong evidence for 13 studies, fairly strong evidence for 3 studies and weaker evidence for 1 study' suggest replacing with '13 studies provided strong evidence, 3 studies...' etc

Authors responses:

We have changed the sentence:

“According to the grading criteria (Table 1) it can be concluded that the included studies differed substantially; they were based on different study designs, contained a variety of outcome measures and varied in quality. 13 references provided strong evidence, three references provided fairly strong evidence and one reference provided weaker evidence.”

Reviewer comment:

p6 line 49 Suggest replacing 'Which support interventions are evaluated that focus on effects for the children?' with 'What types of interventions that focus on effects for the children were evaluated in the studies?'
Authors responses:

We have changed the sentence:

“What types of interventions that focus on effects for the children were evaluated in the studies?”

Reviewer comment:

p7 line 53 Throughout this section and the 'key results' section, check tenses for consistency.

Authors responses:

The native English speaking co-author has checked tenses for consistency in this section and throughout the manuscript.

Reviewer comment:

p8 line 4 'expected respectively' check grammar

Authors responses:

The grammar has been edited accordingly for this sentence. The native English speaking co-author has also checked and edited grammar throughout the manuscript.

Reviewer comment:

p9 line 9 'Support for the parents' - should this be 'feelings of being supported’?

Authors responses:

We have changed the text to:

“parent’s feelings of being supported”
Reviewer comment:

p10 line 23 Please provide reference for this systematic review (presumably Rosner et al)

Authors responses:

We have added the reference to Rosner et al. (2010).

Reviewer comment:

p10 lines 33-34 - define primary and secondary preventative for audiences not familiar with this typology

Authors responses:

We have added a definition:

“It is also important to keep in mind that most of the included interventions were primary or secondary preventive. That is they sought to prevent the development of an illness or disease before it ever occurred or lower the impact if it already had occurred, and thus effect sizes could be expected to be small, but still be of importance for a large group of children.”

Reviewer comment:

p10 line 41-42 Conclusion that interventions need to be directed at both needs a little more support, eg is the effect size for children's outcomes greatest in the interventions that target both parents and children, or is this conclusion being drawn because large effect sizes were found for both groups

Authors responses:

We have changed this sentence:
“One conclusion from this review of interventions is that there are studies that have shown effects for children and their caregivers. The results indicate that supportive interventions can be directed exclusively to the children or to both the bereaved child and the child’s remaining parent or caregiver. Support for the children’s caregivers can strengthen their own health and their capacity to support their children. A supportive parenting is a protective resource for parentally bereaved children” (with reference to Haine et al. 2006).

Reviewer comment:

p10 The paragraph starting 'the article reviews studies' effects' contains a number of points, which could usefully be separated and possibly expanded e.g. support for both/support directed at children/value of group support/value of family support/insufficiency of programmes for some children/

Authors responses:

We have added more details in the results section. The changes are highlighted.

Reviewer comment:

p11 The paragraph starting 'This systematic review' is v important and well written

Authors responses:

Thank you very much!

Reviewer comment:

p11 line 57 Maybe need to add something about whether these groups might be under-represented in evaluation studies
Authors responses:

We have added a sentence about why these children may be under-represented in evaluation studies:

“One explanation why these children are underrepresented in evaluation studies is that the largest proportion of children in the western world live together with both their parents. It is difficult to conduct evaluation studies with this vulnerable group of children.”

Bridget Candy (Reviewer 3):

Reviewer comment:

The authors have provided a potentially informative systematic review. However there are a lot of limitations in their text which reduce the clarity and impact this work could have.

Authors responses:

Thank you for valuable comments.

Reviewer comment:

My recommendations are for them to follow more closely accepted methods for high quality systematic reviews. Such as using the PRISMA guidelines. It may also help to take advice from a colleague familiar with undertaking systematic reviews of trialled interventions and also someone who is a native English speaker as some of the sentences are a little 'clumsy' (such as (page 7) 'it is most common that the parents died die to illness' - a clear way to express this would be 'Commonly parents died because of an illness').

Authors responses:

We have chosen this level of ambition for this article. We have previously followed editor Nancy Prestons’ comments on an earlier version of the article (comments by email 2015-12-18). Preston wrote: “Before we proceed to peer review could you add some more details to your methods and results? You are handling predominantly randomised controlled trials but have not produced any effect sizes for the studies in the tables such as odds ratios or relative risks or indeed how you
analysed and synthesised results except in a narrative manner. Even if you cannot arrive at a cumulative statistic - by adding the same summary statistic where possible to the table would help in the results/key findings section. If you are following Cochrane methods we would have expected effect size reporting and how a summary statistic might have been used.”

One of the authors is a native English speaker.

The sentence is changed (page 7):

“Commonly parents died because of an illness.”

Reviewer comment:

Finer comments on MS are:

ABSTRACT:

In methods section - state first your inclusion criteria. Then where you searched included dates to searched to. Do not have results in this section In results section - provide a statement on number that were trials and provide a statement on quality of trials. Aim to give some statistical evidence of these positive results you present.

Authors responses:

The abstract is revised according to the reviewer’s comments.

Reviewer comment:

MAIN TEXT:

In methods section - Put the first and second search in the same sentence. What are 'Central' search terms? It is usual to put the inclusion criteria first and then state where you searched. Also why did you search beyond RCTs? You found sufficient RCTs not to warrant extending
this and your conclusions to weaker designed studies. Moreover, your message would be clearer and more robust. Why did you chose that particular grading system to assess quality, why did you not use the Cochrane approach? Please be more specific on what you mean by excluding small studies, how small is small, n = 5 or n = 50 ?

Authors responses:

The inclusion criteria are now presented first in the methods section.

The first and second searches are now presented together.

The term “central” is removed.

As there were few RCT studies we also included controlled cohort-studies, since not only RCT:s but even well conducted studies of other design might be contribute to the evidence of a method as is acclaimed for instance in the GRADE system (Atkins D, Best D, Briss PA, Eccles M, Falck-Ytter Y, Flottorp S, et al. Grading quality of evidence and strength of recommendations. BMJ 2004;328:1490, . Guyatt G, Oxman AD, Akl EA, Kunz R, Vist G, Brozek J, et al. GRADE guidelines 1. Introduction – GRADE evidence profiles and summary of finding tables. J Clin Epidemiol 2011;64:383-94), which is used internationally by for example WHO, NICE, Cochrane Collaboration and BMJ.

Grading system: We used a grading system that has previously been accepted in this journal Hudson et al. (2010) A systematic review of psychosocial interventions for family carers of palliative care patients. BMC Palliative Care. 2010;9;17. doi:10.1186/1472-684X-9-17

We have added a sentence in the methods section that we excluded studies with less than 30 participants.

Reviewer comment:

Analysis section in methods

Why did you not consider a meta-analysis of the RCTs, this method is what if possible systematic reviews of interventions aim for. They give a more precise estimate of the effect of trials of sufficient similarity? Presentation is not standard, again they would be wise to consult a systematic reviewer or a statistician. Some of the statistics and presentations are not usually advised by statisticians, such as Yates correction (see Haviland, Mark G. "Yates's correction for continuity and the analysis of 2× 2 contingency tables." Statistics in medicine 9.4 (1990): 363-
3670. Also it is not usual to calculate the statistical power, as an ad hoc calculation after analyses. Another issue is in providing sig as ns, *, ** statistician prefer actual values.

Authors responses:

We have chosen this level of ambition for this article. We have previously followed editor Nancy Preston's comments on an earlier version of the article (comments by email 2015-12-18). Preston wrote: “Before we proceed to peer review could you add some more details to your methods and results? You are handling predominantly randomised controlled trials but have not produced any effect sizes for the studies in the tables such as odds ratios or relative risks or indeed how you analysed and synthesised results except in a narrative manner. Even if you cannot arrive at a cumulative statistic - by adding the same summary statistic where possible to the table would help in the results/key findings section. If you are following Cochrane methods we would have expected effect size reporting and how a summary statistic might have been used.”

We have recalculated the chi-2 tables, without Yates’s correction.

We have also omitted the statistical power column. There is an ongoing debate over the use of calculating statistical power after conducting a trial. It’s not advisable to use it as an explanation of non-significant findings since the power will always be low. However, some authors claim that it might be informative in an independent comparison. However, we agree with the reviewer that it is not common and it might blur the picture, thus we only report effect seize in addition to actual values to significance.

Reviewer comment:

Results sections

Please move section on study design to below your first two paragraphs in results. In the section on included studies first sentence, the second paragraph this would be better placed in the methods section than results.

Authors responses:

We have placed the section study design in the results section as suggested.

Table 3 and table 4 have switched headings.
The text about analysis is now placed in the methods section “Data analysis”.

Reviewer comment:

Please add a separate section on quality assessment and reconsider use of Cochrane system.

Authors responses:

We have added a separate section on quality of the included studies. However we still use the grading system. We used a grading system that has previously been accepted in this journal Hudson et al. (2010) A systematic review of psychosocial interventions for family carers of palliative care patients. BMC Palliative Care. 2010;9;17. doi:10.1186/1472-684X-9-17

Reviewer comment:

Please relabel section 'key results of the included interventions' to 'effectiveness of interventions'. This makes it more precise for the reader. I don't understand why you have lumped together all the results from the trials with the weaker designed studies. I would present the results separately. Could any of these results be combined across RCTs?

Authors responses:

The heading of the section is changed to “Effectiveness of interventions”.

We have added information about the evidence of the studies in accordance with the grading criteria. We have also changed the presentation into three sub-headings: large effects, medium effects and small effects. We consider that it is now easier for the reader to follow which studies have strong evidence and large effects, strong evidence and medium effects, strong evidence and small effects and so on.
Reviewer comment:

DISCUSSION

I think they need to rewrite the discussion to make it clearer for the reader. The sections I would like to see are those seen in Cochrane systematic reviews:

1-What the review set out to do.

2-A summary of the key findings

3-Overall completeness and applicability of evidence

4-Quality of the evidence

5-Potential bias in the review process

6-Agreements and disagreements with other studies or reviews

7-Implications for practice

8-Implications for research

Authors responses:

The discussion is rewritten. We hope this will make it clearer for the reader.

Linda Oostendorp (Reviewer 4):
Reviewer comment:

Thanks for inviting me to review this systematic review on bereavement interventions for parentally bereaved children. This review is generally well performed and the manuscript is well written, but could benefit from a more detailed description of study methods and a more insightful way of presenting the results. My detailed comments are listed below.

Authors responses:

Thank you very much for valuable comments.

Reviewer comment:

Abstract

The abstract does not follow PRISMA guidelines for reporting of systematic reviews, e.g. data sources, study appraisal and synthesis methods are not described in the abstract.

Authors responses:

Abstract is rewritten. We have added information about sources, study appraisal and analysis.

Reviewer comment:

Methods:

Page 4: Search strategy

It would be helpful if the authors could include the search strategy in an Appendix.

It is unclear to me why the authors decided to use a different search strategy when updating the search in November 2015, rather than repeating the search strategy used in April 2013 to identify new publications.
Authors responses:

The literature search in 2013 was conducted with the help from an information specialist at the National Board of Health and Welfare Sweden (NBHWS). In 2015 we wanted to update the literature search. Unfortunately at this time we could not get help from NBHW. Instead we had assistance from a librarian at Linnaeus University who used a slightly different strategy for the literature search. However, the thesaurus/mesh-terms/headings were the same for both searches. One of the databases PILOTS was not available at Linnaeus University.

Reviewer comment:

Page 5

Selection criteria: The authors excluded studies where the population was too small for the results to be generalisable. Could the authors please provide more details.

Authors responses:

We have added a sentence in the Method section that we excluded:

“Studies with a population of less than 30 participants”.

Reviewer comment:

Data analysis: it would be helpful if the authors could add an explanation of the Common Language Effect size, which readers may not be familiar with.

Authors responses:

We have added an explanation. See also our previous response to Reviewer 1 above.
Reviewer comment:

Results:

Table 2: this Table presents details for 12 studies, where I would have expected 17 studies. Could you please clarify why 5 studies (references 29, 32, 40, 41, 43) are missing from this Table?

The Table shows that numbers were missing or could not be calculated from some of the original studies. Did the authors attempt to contact the authors of these studies to obtain these data?

Authors responses:

We have added an explanation about the references that are not presented in the Table 2:

“There are 12 references that analyse effects within and between groups, while five references analyse moderating and mediating factors. The latter are excluded from the analysis of effects in the table 2, but are nevertheless informative and are therefore included in the article.”

We have not contacted the authors for missing data.

Reviewer comment:

Table 3: The study of Kalantari et al. 2012 seems out of place in this review. The population is quite different from the other studies included in this review, since the refugee children had lost both parents as well as other family, their home and school and were traumatized by war and living as a refugee in a foreign country.

Authors responses:

We have chosen to include the study of Kalantari et al. (2012) because there are currently a lot of refugee children and unaccompanied refugee children in Sweden and in other countries in Europe. Many of these children have experienced a loss of a parent/parents and are in need of support. We consider it is important to gain knowledge about effective support even for this group of parentally bereaved children.
Reviewer comment:

Page 8-9: Key results of the included interventions are presented by effect size which I find a bit confusing. I would suggest presenting results by type of outcome as presented in Table 4. Many of the studies showed non-significant differences between treatment and control groups (Table 2) and this is currently not reflected in the description of results.

For example, for Children's health and behaviour, the most common outcome measures were the Child Behaviour Checklist (CBCL), Children's Depression Inventory (CDI), Youth Self-Report (YSR), and Children's Manifest Anxiety Scale-Revised (R-CMAS) as shown in Table 4. According to Table 2, 25 comparisons were available (12 using the CBCL, 5 using the CDI, 4 using the YSR and 4 using the CMAS-R), of which only one showed a significant effect between groups.

Authors responses:

As there are a lot of measures in the included studies (50) we have added the following summarizing sentence in the results section:

“There are positive effects from interventions for children’s health, self-esteem, grief symptoms, behaviour problems, school problems as well as for caregivers’ parenting and mental health.”

We have also added a sentence in the results section that there are studies that have not shown effects on some measures.

Reviewer comment:

In a similar vein, Table 2 shows that studies evaluating the Family Bereavement Program have showed many non-significant differences between the treatment and control groups, which is not reflected in the results section.
Authors responses:

We have added a sentence in the results section that there are studies that have not shown effects on some measures:

“There are also included studies that did not show effects on some measures for example for anxiety, depression, internalizing and externalizing for the subgroup boys. This result indicates a need to pay attention to possible gender differences. It is also important to note that most of the studies in the review are underpowered, thus indicating that there is a risk that there in some cases might actually have been a difference between the intervention and control group, which may not have been detected due to the fact of the samples being too small to find statistically significant differences when the effect sizes are small.”

Reviewer comment:

Discussion:

An important gap in research identified in this review but not addressed by the authors is that all interventions, apart from 'Writing for Recovery' are English language interventions evaluated in the UK or USA.

Authors responses:

In the section “Implications for research” we have added a sentence that:

“All references, apart from one, are studies about English language interventions evaluated in the USA or UK.”

Reviewer comment:

The authors' conclusion that bereavement interventions should be directed at both the child and the caregiver does not seem to follow logically from the data presented in the results section.
Authors responses:

We have changed this sentence to the following:

“There are interventions directed exclusively to the children or to both the bereaved child and the child’s remaining parent or caregiver. Included studies have shown effects for both children and caregivers.”

Reviewer comment:

The authors suggest conducting further studies among younger children, potentially using qualitative interviews to evaluate interventions. However, such studies would have been excluded from the current review as studies were required to offer quantitative outcome measures. Qualitative data from evaluations could also be helpful to identify opportunities to improve current bereavement interventions.

Authors responses:

We have added the following sentence in the results section “implications for research”:

“Qualitative data from evaluations could also be helpful to identify opportunities to improve current bereavement interventions.”