Author’s response to reviews

Title: Physical and psychosomatic health outcomes in people bereaved by suicide compared to people bereaved by other modes of death: a systematic review

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Author’s response to reviews:

Following submission of the original manuscript (on 14/12/2017) the authors became aware of one additional paper (Erlangsen et al 2017) that met the inclusion criteria for the systematic review, which was published on 22/03/2017. Following consultation with the managing editor, we are advised to include this additional paper and any other paper(s) that may be identified. Upon re-running of the searches, this was the only new paper identified. This paper has now been incorporated into the systematic review. Therefore, please be aware that the total number of included studies has increased from 23 to 24. The newly located paper found that those bereaved by suicide had a lower risk of a number of physical health disorders, including cancers, diabetes, cardiovascular and chronic lower respiratory tract disorders compared to those bereaved by other causes of death. The addition of this paper has not altered the conclusions originally drawn.

We would like to thank both of the reviewers for taking the time to review this manuscript and for their helpful comments and suggestions. Please find our responses to your comments below:

Reviews

“I recommend some minor revisions to the discussion as follows:
In the discussion, it is not clear what is meant by "kinship" (Line 383):

Therefore, it appears that kinship may be associated with the risk of adverse outcomes following suicide bereavement.

Are you describing the difference in impact between children and adults by using the term "kinship" if yes this needs to be described in more detail”.

Response: The authors acknowledge that the term kinship should be explained in the text. Kinship refers to the type of familial relationship (parent-child, spousal, sibling, child-parent), including blood and non-blood relationships, between two people. In short, kinship is referring to the type of familial relationship between the deceased and the surviving family member. This explanation has been incorporated into the text to clarify the meaning of kinship (Results section, page 18).

Line 392 - use of the term "death" would it be more appropriate to use "suicide"

Response: The authors acknowledge that it is more appropriate to use “suicide” rather than “death”. The text has been changed as followed to reflect this, as follows (Discussion, page 21):

“Of the six significant studies, two examined parents bereaved by the suicide of their offspring [19, 31]. The remaining studies examined children bereaved by parental suicide [33], suicide-bereaved relatives and spouses [30], bereaved spouses [34] and bereaved spouses and parents [32]”.

The first two paragraphs of the discussion are far too long and both extend over 1 page each - these need to be confined to smaller paragraphs to make the meaning of each clearer

Response: The first two paragraphs have now been broken up into two in order to improve clarity (Discussion section, page 20)
The current systematic review of physical and psychosomatic outcomes found tentative evidence supporting an association between bereavement by suicide and some physical health outcomes, although there are inconsistencies. Cardiovascular disease, COPD, hypertension, diabetes, increased pain and poorer general health were some of the adverse physical health outcomes noted in people bereaved by suicide [19, 30-32]. However, a large number of studies found no significant differences or found that suicide bereavement conferred a lower risk of various physical and psychosomatic health outcomes [20, 22-24, 26, 33-44, 46, 47].

Of the six studies that found a positive association [31, 32], three had large sample sizes which would increase the likelihood of finding a positive association if an association exists [19, 30, 34]. Two studies found an association in the opposite direction. One of the studies found that those bereaved by suicide had a lower risk of a number of symptoms and illnesses, including cancers, diabetes, cardiovascular and chronic lower respiratory tract disorders compared to those bereaved by other causes of death [34]. The other study found that non-suicide bereaved children visited a GP more frequently than suicide-bereaved children [33]. Study design varied across these studies, and included one case-control, two cross-sectional and three cohort studies. Two of the studies were conducted in Canada with the remaining four studies being conducted in the United States, The Netherlands, Denmark and Japan [19, 30-32, 34]. Only one of the studies revealing significant results, adjusted for pre-bereavement functioning of the bereaved [19].

Line 414 - shame and stigma, this should be referenced and it would be good to develop this explanation further. I recommend a short paragraph on this - there is a large body of evidence linking shame/stigma and poor health outcomes, and this is highly relevant to suicide bereavement.

Response: The authors agree with the reviewer and have included appropriate references and a further explanation of stigma/shame and poor health outcomes as follows (Discussion section, page 23-24).

"Research indicates that family and friends bereaved by suicide experience more perceived stigma and shame than those bereaved by both sudden unnatural and sudden natural death [51]. Stigma has been linked to a number of avoidance behaviours, including concealing the death and social withdrawal [52, 53]. People bereaved by sudden death, including those who perceive that they are highly stigmatised, are at greater risk of suicidal behaviour and depression [54]. In addition, a recent systematic review highlighted that stigma experienced by people bereaved by
suicide was highly correlated with increased somatic reactions, including headaches and stomach pain [52]. Research has moreover posited that that shame and stigma may be a key component related to poorer helpseeking in people bereaved by suicide [51, 52]. Consistent with this, one study we located found that somatic hospital use was less for women bereaved by a spouse’s suicide, while men were less likely to visit a GP than men bereaved by other causes of death [34]. Interestingly, suicide-bereaved spouses had a higher risk of dying by any cause compared to people bereaved by the non-suicide death of a spouse. The authors of this paper therefore allude that suicide-bereaved spouses may be less likely to seek medical care for health concerns due to preoccupation with grief”.

After the limitations and strengths, please describe the implications of this review - what specifically does it mean for future research? Be more explicit

Response: Please see below for this addition to the text (Discussion section, page 26-27):

“The evidence indicates that suicide bereavement may be positively associated with a number of adverse physical health outcomes but there was disagreement across the studies. In addition, studies included relating to psychosomatic health outcomes did not show an association with suicide bereavement. Some of the papers investigating psychosomatic health outcomes had small sample sizes, selection bias issues and failing to control for confounding factors. Therefore, further research is necessary to address the uncertainty regarding the association between physical and psychosomatic health outcomes and suicide bereavement. Register-based and cohort studies represent the most appropriate designs to address this research question. The importance of selecting an appropriate control group, such as people bereaved by sudden and violent deaths, cannot be overemphasised. Future studies also need to allow for sufficient time to follow-up as some of the outcomes may not be present shortly after bereavement”.

This manuscript has a clear and informative title. It reports a systematic review on people bereaved by suicide compered to those bereaved by other kinds of death. Well conducted, need some writing revision. It approaches an important issue, however it needs some improvement in discussion, leaving the article with a lot of data report but a lack of data interpretation, and poor in a Take Home Massage.
1. Introduction:

• I suggest a revision of the fourth paragraph. It seems a bit long and all the explanation from “Several explanations” to “conceal the cause of death” brings a confusing scenario.

Response: The authors acknowledge that this paragraph could be improved upon and have now added further research findings to this paragraph to improve the clarity, as follows (Introduction section, page 4-5):

“Following on from this, people bereaved by suicide are at increased risk of engaging in suicidal behaviour themselves [11]. Several explanations have been posited to explain why those bereaved by suicide are a particularly at-risk bereaved group, in terms of genetic, social, psychological and physical vulnerabilities. Firstly, it has been established that suicidal behaviour is familial and may be partly explained by genetics [12, 13]. Research indicates that proband suicide attempt increased the odds of offspring suicide attempt by nearly 5-fold, even when mood disorder at the time before the suicide attempt, baseline history of suicide attempt and mood disorder were controlled for [12]. Some research suggests that the intrafamilial transmission of impulsive aggression, childhood maltreatment and mood disorder may be possible mediators [12]. However, the exact mechanism underlying this genetic transmission is still unclear”.

• The aim should be addressed in this part of the manuscript, as it was done in the abstract, and may be very well emphasized at the last paragraph.

Response: The authors agree: the aim has now been included as follows (Introduction section, page 5):

“Therefore, it is also critical to consider somatic and psychosomatic symptoms and complaints which may be more likely to present in the short-term following bereavement [21-24].

However, a synthesis of research on the effects of suicide bereavement on physical health problems and psychosomatic symptoms has not been conducted to date. The rationale for this review is to contribute to the evidence around the societal burden of suicide bereavement as
borne by the families and health services, as well as informing clinicians who support those bereaved by suicide. The aim of this paper is to examine and compare the physical and psychosomatic morbidities of people bereaved by a family member’s suicide with family members bereaved by other modes of death”.

2. Methods:

• Maybe the first paragraph of Methods should be part of the introduction section, starting this part of the manuscript by the actual second paragraph.

Response: The authors agree with the reviewer that the first paragraph of the methods would be better placed in the last paragraph of the introduction. This has been incorporated as follows (Introduction section, page 5):

“However, a synthesis of research on the effects of suicide bereavement on physical health problems and psychosomatic symptoms has not been conducted to date. The rationale for this review is to contribute to the evidence around the societal cost of suicide bereavement as borne by the families and health services, as well as informing clinicians who support those bereaved by suicide. The aim of this paper is to examine and compare the physical and psychosomatic morbidities of people bereaved by a family member’s suicide death with family members bereaved by other modes of death”.

• Was minimum sample size calculated for representativeness?

Response: Minimum sample size was not calculated for representativeness as representativeness is not necessarily reflected by sample size. Given the dearth of research in this area, all relevant studies were included regardless of size. However, the quality rating of included studies took into account the issue of sample size/representativeness.
• I suggest reviewing the Methodology presentation to become more clear and enlightening. The research procedure seems a bit unclear explained and replicability of this study uncertain.

Response: The authors have presented the information regarding the methodology more clearly. This has been done by providing the PROSPERO registration number of the review, the search strategy, including databases searched, search terms and any limits applied, inclusion and exclusion criteria, list of items extracted from the articles and chosen risk of bias assessment tool.

• Risk of bias assessment is addressed only in results section. It’s approach is a core part of a Systematic Review Structure.

Response: The authors acknowledge that the inclusion of the risk of bias assessment was unclear. A new heading has been included in the methods to highlight the risk of bias assessment to improve clarity for the reader (Methods section, page 9).

3. Results:

• The representativeness of 23 papers instead 26 studies should be clearly explained.

Response: The authors agree that the explanation given was unclear and have revised this section as follows (Results section, page 10):

“...A total of 6,959 records were identified across the four databases, with four additional records identified from other sources, namely reference list searching. Eighty-six full-text articles were assessed for eligibility. The search was re-run in March 2017 which retrieved 666 articles that were published in the interim. One of these met the criteria for the study. Therefore, 24 papers meeting the inclusion criteria, representing 27 studies were included in the review. Three papers were published using the same study sample. Where this occurred, the most up-to-date or most...
comprehensive information and results were included. This was done to ensure that information was not duplicated in the review”.

• Since selection bias may be an issue on some studies, refusals individuals were described? Shouldn’t the description be approach?

Response: The authors are unclear regarding the meaning of the reviewers comment above. If the reviewer is asking about procedures for recruitment and attrition within each of the included studies, the authors acknowledge that this is very important from a methodological viewpoint and this has been discussed in the risk of bias assessment paragraph (Results section, page 11-12). The issue of self-selection bias has also now been addressed in the risk of bias assessment paragraph and also in the results section as follows (Results section, page 12-13):

“A comparison of characteristics of responders and non-responders was present in a minority of the studies (2/24), with the majority of papers not presenting this information (13/24). Six studies were register-based studies and therefore, the issue of non-response bias is not applicable. One study did not have any information on non-responders beyond gender, age, mode of death, and place of residence of deceased due to confidentiality reasons. A further study compared excluded cases to remaining cases on victim’s age, race, sex and method of death and concluded there was no evidence of sample bias. Finally, one study compared bereaved offspring that remained in the study to those lost to follow-up. Bereaved offspring lost to follow-up were more likely than those who remained in the study to have a caregiver with a history of alcohol or substance disorder (32.1% vs. 16.7%), to have a caregiver of minority status (28.4% vs. 11.7%), and to have had a proband with a history of an anxiety disorder (28.3% vs. 16.4%). Overall, selection bias emerged as an important methodological consideration in the included papers”.

• Once no-bereaved controls were exclusion criteria, why describe While Wilcox and colleagues study in the results? Shouldn’t be better addressed only at discussion?

Response: The authors agree with the reviewer and this paper has now been removed from the results section.
4. Discussion:

- Did the authors understand that the included studies were overall reliable?

Response: Overall, the included study types in this review are reliable. However, the authors accept that there were varying levels of quality across the studies. These issues are described in detail in the risk of bias assessment section and also throughout the results and discussion section.

- Is it really possible to make the statement that there are associations between bereavement by suicide and physical health outcomes, since “there are inconsistence” and the studies had different designs for a meta-analysis? The way it’s written seems more like the authors opinion than evidence based information. I suggest approach revision, even more after reading the first two lines of conclusion section.

Response: The statement that the reviewer is referring to is as follows:

“The current systematic review of physical and psychosomatic outcomes found that bereavement by suicide is associated with some physical health outcomes, although there are inconsistencies”

The authors have now reworded this sentence and added in further sentences to reflect the uncertainty in arriving at a definitive conclusion regarding the association between suicide bereavement and adverse physical health outcomes, as follows (Discussion section, page 20):

“The current systematic review of physical and psychosomatic outcomes found tentative evidence supporting an association between bereavement by suicide and some physical health outcomes, although there are inconsistencies. Cardiovascular disease, COPD, hypertension, diabetes, increased pain and poorer general health were some of the adverse physical health outcomes noted in people bereaved by suicide [19, 30-32] compared to those who experienced other types of bereavement. However, a large number of studies found no significant differences or found that suicide bereavement conferred a lower risk of various physical and psychosomatic health outcomes [20, 22-24, 26, 33-44, 46, 47]”.
The authors acknowledge that a meta-analysis is not suitable due to the heterogeneity in outcomes of included studies and study designs therefore, a narrative synthesis approach was chosen. Other similar papers in the area of suicidology have also used narrative synthesis in these circumstances, including Pitman et al (2014) and Sveen and Walby (2008).

• Review Discussion section, it brings some repetitive information (addressed before in Results Section).

Response: We thank the reviewer for bringing this to the attention of the authors. The discussion section has been revised to ensure that information has not been repeated from the results section. Further literature has also been added to improve clarity as follows:

“Research indicates that family and friends bereaved by suicide experience more perceived stigma and shame than those bereaved by both sudden unnatural and sudden natural death [51]. Stigma has been linked to a number of avoidance behaviours, including concealing the death and social withdrawal [52, 53]. People bereaved by sudden death, including those who perceive that they are highly stigmatised, are at greater risk of suicidal behaviour and depression [54]. In addition, a recent systematic review highlighted that stigma experienced by people bereaved by suicide was highly correlated with increased somatic reactions, including headaches and stomach pain [52]. Research has moreover posited that that shame and stigma may be a key component related to poorer helpseeking in people bereaved by suicide [51, 52]. Consistent with this, one study we located found that somatic hospital use was less for women bereaved by a spouse’s suicide, while men were less likely to visit a GP than men bereaved by other causes of death [34]. Interestingly, suicide-bereaved spouses had a higher risk of dying by any cause compared to people bereaved by the non-suicide death of a spouse. The authors of this paper therefore allude that suicide-bereaved spouses may be less likely to seek medical care for health concerns due to preoccupation with grief” (Discussion section, page 24).

“This is the first review to synthesise all relevant papers related to suicide bereavement and physical and psychosomatic health outcomes. The evidence indicates that suicide bereavement may be positively associated with some adverse physical health outcomes, but there was dissonance across the studies. Studies relating to psychosomatic health outcomes failed to find an association with suicide bereavement. Considering the relatively small number of studies and small sample sizes, it is therefore recommended to conduct further research addressing the
uncertainty regarding the association between physical and psychosomatic health outcomes and suicide bereavement. Register-based and cohort studies represent the best study designs to examine this research question. The importance of selecting an appropriate control group, in this case, other people bereaved by sudden and violent deaths, cannot be overemphasised. Future studies also need to allow for sufficient time to follow-up as some of the outcomes may not be present shortly after bereavement” (Discussion section, page 26-27).

• Limitations should be approach in a specific Limitation Section.

Response: The authors agree with this point raised by the reviewer. There is now a specific heading given to the strengths and limitations of the review (Discussion section, page 26-27).

5. Overall Issues:
• English language is clear and understandable;

• Absence of a clear Take Home Message.

Response: The take home message has now been improved upon, as in the conclusion section as follows (Conclusion section, page 27-28):

“This systematic review found that a small number of studies demonstrated associations between suicide bereavement and adverse physical health outcomes, including cardiovascular disease, diabetes, chronic obstructive pulmonary disease, hypertension and poorer general health. However, most studies failed to conclude that people bereaved by suicide were at higher risk for a number of physical health conditions compared to non-suicide bereaved individuals. No studies found a significant association between suicide bereavement and psychosomatic health outcomes. Thus, findings of this review indicate that there are more similarities than differences between people bereaved by suicide and people bereaved by other causes of death. Inconsistencies in results may be due to methodological shortcomings in the available studies, including inappropriate selection of control groups, small sample size and failure to control for confounding factors. Furthermore, there is some evidence indicating that risk of physical health
outcomes may differ according to familial relationship. Further longitudinal controlled studies need to be conducted in order to better understand the health implications of suicide bereavement, specifically compared to other types of bereavement after sudden and violent deaths, including accident and homicide deaths.”