PEER REVIEW HISTORY

ARTICLE DETAILS

TITLE (PROVISIONAL)	Healthcare consumption in parents of young terrorism survivors: A registry-based study in Norway
AUTHORS	Haga, Jon Magnus; Thoresen, Siri; Stene, Lise; Wentzel-Larsen, Tore; Dyb, Grete

VERSION 1 – REVIEW

REVIEWER	Vanessa Cobham The University of Queensland Australia
REVIEW RETURNED	20-Jul-2017

GENERAL COMMENTS	The authors are to be commended on this work, which makes use of a national registry in order to examine pre and post-disaster health care consumption among parents of youth involved in a terrorist attack. This work emphasises the importance of thinking about 'survivors' of a potentially traumatic event in a broader, more systemic way than is typical. For the most part, my comments are minor, however there are a couple of things that I think it would be helpful for the authors to clarify and/or elaborate upon in a revision. 1. In the Abstract, and in the Discussion, there is a tendency to generalize the results of this study to other types of potentially traumatic event exposures (particularly natural disasters). Based on the literature reviewed in this paper, this generalization does not seem warranted (indeed, in the Discussion, contrasting findings are described in a study conducted with Danish survivors of the 2004 tsunami - where specialized mental health consumption was associated with history of service consumption rather than disaster-related distress). Thus, for example, I would suggest changing the conclusion sentence in the Abstract to refer specifically to 'terrorist disasters' rather than disasters more generally. 2. Page 5, paragraph 3: "Post-disaster, healthcare resources may be scarce". I found this sentence to be a little confusing. Presumably the authors are referring to a scarcity in healthcare resources for dealing with post-disaster mental health care issues? 3. It was interesting to read about the dedicated contact people who were in place to monitor and support families in the first year, and facilitate access to healthcare services (pp. 6-7). As I was reading the paper, I wondered, what impact (if any), this would have on healthcare service consumption. In the Discussion, there is a statement on p.

	14 about 'psychosocial support through the outreach program'. I would have liked to have seen a bit more information provided about this - were the contact people part of this program? Without any information about what this program involved, it was difficult to know how this program might have alleviated healthcare needs (as well as potentially increasing demand through assisting people to seek help). Further elaboration would be useful. It might also be worth mentioning in the Discussion the extent to which this kind of outreach program (whatever it did involve) is typical in different parts of the world - and the extent to which the results found in this study and this context are generalizable. 4. Could the authors please elaborate a little on what is involved in the somatic and mental healthcare services? 5. In the Method, under the Statistics heading, I was unclear about the 'observational period' discussed in the last paragraph of p. 9. 6. I was also unclear on the role of the predictions regarding healthcare use (given that the actual data were available for the relevant time points). Or were the predictive values used only for the post-disaster time points perhaps (i.e., to predict what the pattern of usage would have been if there had been no attack)? The age used for the predictions (47 years) didn't seem to be explained - it is a little above the average age of mothers in the sample and just under 3 years below the average age of fathers in the sample. 7. I wasn't clear on whether participants' pre-disaster healthcare consumption was controlled for in this study. This is clearly important in light of the study of Danish survivors reviewed in the Discussion. Could the authors please clarify this. In relation to the study of Danish survivors of the 2004 tsunami, it is interesting to speculate on the potential differences of being impacted by a natural disaster (typically - though not always - seen as an act of nature or God) vs. a man-made disaster such as the attack which is the index trauma in this paper. 8. In relation to the last point above, and the lack of literature in this area, I would suggest that the authors are even more tentative in their statement (p. 14) that similar patterns of healthcare need may ensue after other traumatic exposures of parents. Also, in relation to this statement, it was unclear whether the authors were talking about potentially traumatic events that directly impact only children. I would like to congratulate the authors on this interesting, clearly presented piece of research which makes an important contribution to the literature. Thank you for the opportunity to review this paper.

REVIEWER	Sarah Halligan University of Bath No Competing Interest
REVIEW RETURNED	01-Aug-2017

GENERAL COMMENTS	This manuscript provides important information about health service usage among parents of children exposed to a terror attack. As the authors point out, this severe event counted as a Criterion A trauma for parents as well as children, but indirect exposure to trauma in parents is often overlooked. The current manuscript provides a compelling case for the need to consider prolonged psychological distress following such an exposure in parents. I have the following minor comments. 1. There are many grammatical errors that need addressing throughout. Sometimes these result in errors in reporting (e.g., on page 13 – "only a majority of parents" should be a minority). This needs careful attention in any revision. 2. In the methods, please can the authors provide brief summary of their original sample, versus simply referring to another publication, so that the extent to which was representative of the relevant parent population can easily be ascertained? 3. The statistical treatment needs more explanation in order to be transparent for a non-expert reader, particularly the choice of regression model. To what extent do statistical analyses take account of time sensitive information presented in detail in the second figure, versus simply combine data over periods specified? Clarifying the link between figures and key statistical analyses would be helpful - as far as I can tell analyses are represented by Figure 2, with other data being descriptive, but I wasn't certain. Several specific points also need clarification. What were the results of analyses based on only in person consultations (referred to as being in supplementary material, but there are no data that duplicate Figure 2 and results are not clear). What does it mean that 'improved quality of the NPR registry during the pre-disaster period was observed' and what did associated sensitivity analyses show? 4. Tables/figures need improvement in order to be accessible. Figure 2, which seems to provide the key information, is difficult to process and I wonder whether some of this material could simply have been tabulated. Overall, figures seemed overly complex, need further annotation in order to be completely clear. It currently isn't clear what distinct, essential information each figure provides. The text refers to colours in violin figures that were not represented in the manuscript. Supplementary tables are even less accessible, and not all are referred to in the text so it wasn't clear what information was intended to be extracted from each. 5. In the discussion, gender differences might be further unpacked. In particular, more in depth consideration of whether this can be interpreted as due to a difference in actual rates of mental health problems or a difference in rates of treatment access would be helpful. It might also be useful if any brief reference to the potential financial burden of the increased healthcare usage could be made.

VERSION 1 – AUTHOR RESPONSE

Reviewer #1

The authors are to be commended on this work, which makes use of a national registry in order to examine pre and post-disaster health care consumption among parents of youth involved in a terrorist attack. This work emphasises the importance of thinking about 'survivors' of a potentially traumatic event in a broader, more systemic way than is typical.

Thank you for this comment. We think that the latter sentence is a very good way of summarizing the implications of this study, and have chosen to include the sentence in the conclusion (p15) of our paper: “Our study emphasizes the importance of thinking about survivors in a broader, more systemic way.”

For the most part, my comments are minor, however there are a couple of things that I think it would be helpful for the authors to clarify and/or elaborate upon in a revision.

Comemnt 1. In the Abstract, and in the Discussion, there is a tendency to generalize the results of this study to other types of potentially traumatic event exposures (particularly natural disasters). Based on the literature reviewed in this paper, this generalization does not seem warranted (indeed, in the Discussion, contrasting findings are described in a study conducted with Danish survivors of the 2004 tsunami - where specialized mental health consumption was associated with history of service consumption rather than disaster-related distress). Thus, for example, I would suggest changing the conclusion sentence in the Abstract to refer specifically to 'terrorist disasters' rather than disasters more generally.

Response: We fully agree with this comment by reviewer #1. The title, abstract and the discussion have been changed accordingly. Regarding the Danish study, please refer to comment #7.

Comment 2. Page 5, paragraph 3: "Post-disaster, healthcare resources may be scarce". I found this sentence to be a little confusing. Presumably the authors are referring to a scarcity in healthcare resources for dealing with post-disaster mental health care issues?

Response: We fully agree that the sentence was confusing. The sentence has been deleted, as a part of the revision of the introduction called for in comment 3 and 4.

Comment 3. It was interesting to read about the dedicated contact people who were in place to monitor and support families in the first year, and facilitate access to healthcare services (pp. 6-7). As I was reading the paper, I wondered, what impact (if any), this would have on healthcare service consumption. In the Discussion, there is a statement on p. 14 about 'psychosocial support through the outreach program'. I would have liked to have seen a bit more information provided about this - were the contact people part of this program? Without any information about what this program involved, it was difficult to know how this program might have alleviated healthcare needs (as well as potentially increasing demand through assisting people to seek help). Further elaboration would be useful. It might also be worth mentioning in the Discussion the extent to which this kind of outreach program (whatever it did involve) is typical in different parts of the world - and the extent to which the results found in this study and this context are generalizable.

Response: We thank reviewer #1 for this comment. We have now included a more detailed description of the outreach program in Methods (pp6-7): “Soon after the terrorist attack, an early proactive outreach program was established, in line with the current trauma-informed consensus of expert opinions (31). Multi-disciplinary crisis teams, established in affected municipalities (n=128), were to provide the immediate psychosocial support to those in need; dedicated contact persons were to proactively monitor and support the affected families throughout the first year post-disaster, and to facilitate access to regular healthcare services as required (28, 32).”

Regarding the comment on whether the early outreach program might have alleviated healthcare needs, as well as potentially increased the demand through assisting people to seek help, and whether such programs are typical in different parts of the world, we have revised the discussion (p15): “In the aftermath of a terrorist attack, many countries are likely to adopt some kind of crisis response, as was reported following the major terror attacks in France (46) and the UK (47). The organization and contents of such programs vary across different country settings (48). Psychosocial support through post-disaster outreach programs may facilitate access to healthcare services and thus potentially increase overall healthcare consumption. On the other hand, it may alleviate health complaints, and thus reduce consumption. Following the Utøya terrorist attack, a majority of the mothers and fathers were contacted by the outreach services in their municipality (28). The contents of the services provided are not known in detail. No adjustment for engagement with the outreach program was made.”

Comment 4. Could the authors please elaborate a little on what is involved in the somatic and mental healthcare services?

Response: We agree that the paper could benefit from more information on somatic versus mental healthcare services. Thus, in the methods, the description of the healthcare services has been revised: “Post-disaster healthcare services were provided by the regular two-level healthcare system in Norway (33). Level one, including the primary healthcare services of general practitioners (GPs) and emergency primary healthcare, provides services for both somatic and mental health complaints and acts as the entry point and gatekeeper to secondary healthcare services. Level two, the secondary healthcare services, provides specialized mental and specialized somatic healthcare services and is accessed through medical referrals only.”

Comment 5. In the Method, under the Statistics heading, I was unclear about the 'observational period' discussed in the last paragraph of p. 9.

Response: In response to this and other comments on the statistics, the section has been rewritten (pp9-10). Observational period is currently described in the following way: “Negative binomial hurdle regressions were chosen for rate predictions, as this method is suitable for overdispersed count data exhibiting excess zeros (39). In order for results to be interpreted in terms of rates, the regressions need to be offset for the observational period, also known as the persontime at risk. In our material, hospital admissions were considered to make an individual unavailable for healthcare services by other healthcare providers. Thus, in our study observational periods were defined as days of non-hospitalization within each time period being investigated.”

Comment 6. I was also unclear on the role of the predictions regarding healthcare use (given that the actual data were available for the relevant time points). Or were the predictive values used only for the post-disaster time points perhaps (i.e., to predict what the pattern of usage would have been if there had been no attack)? The age used for the predictions (47 years) didn't seem to be explained - it is a little above the average age of mothers in the sample and just under 3 years below the average age of fathers in the sample.

Response: Predictions were made in order to adjust for the age in the parent sample (which may influence healthcare consumption). However, we fully agree with the reviewer that an alternative approach would have been to compare pre- and post-disaster values directly. The statistics (pp9-10) has been rewritten in order to clarify this: “The statistical analyses included in this paper compared post-disaster by pre-disaster healthcare consumption. As the parents’ age was hypothesized to influence healthcare needs independently of the terrorist attack being investigated, rates of pre- and post-disaster healthcare consumption were predicted from age-adjusted regression models, rather than assessing the observed pre- and post-disaster values directly. Negative binomial hurdle regressions were chosen for rate predictions, as this method is suitable for overdispersed count data exhibiting excess zeros (39).”

In order to address the reviewer’s comment on the age used for the predictions, the following has been added: “Negative binominal hurdle regression is a two-component regression model (39). Thus, model predictions need to be calculated for a predefined index individual. In our study, the index parent was defined as a mother or a father of 47 years of age at the time of the attack, reflecting the median age of the full parent sample. In order to increase comparability, we chose to make predictions for the same age in both mothers and fathers, despite the fathers being slightly older than the mothers.”

Comment 7. I wasn't clear on whether participants' pre-disaster healthcare consumption was controlled for in this study. This is clearly important in light of the study of Danish survivors reviewed in the Discussion. Could the authors please clarify this.

Response: All analyses reported are ratios (after vs before). Thus, regressions are not adjusted for pre-disaster values, but rather directly compared to the pre-disaster value. In order to clarify this, aims has been rewritten: “Separately in mothers and fathers, we investigated whether the frequency of parents’ primary and specialized healthcare service consumption in the early (0-6 months) and delayed (>6-36 months) aftermath of the terrorist attack was higher than in the three-year period before the terrorist attack. Secondly, we examined whether the semiannual proportions of mothers and fathers consuming one or more healthcare services were increased early and delayed, when compared to pre-disaster levels.”

The statistics section has been rewritten, and currently contains the following sentences: “The statistical analyses included in this paper compare post-disaster by pre-disaster healthcare consumption.” and “Finally, post- versus pre-disaster rate ratios (RR) were computed by dividing the rates of healthcare consumption (both the frequency of services consumption and the proportion of individuals accessing services) in the early and delayed aftermath by the corresponding pre-disaster rates”. Further changes to the statistics are outlined in the response to comment #6.

The results section has been revised (p11). Figure 3a-b (which presents the results of the statistical analyses) is currently introduced in the following way: “Figure 3a-b presents the output of the statistical analyses comparing pre- and post-disaster healthcare consumption.”

In relation to the study of Danish survivors of the 2004 tsunami, it is interesting to speculate on the potential differences of being impacted by a natural disaster (typically - though not always - seen as an act of nature or God) vs. a man-made disaster such as the attack which is the index trauma in this paper.
Concerning the comparison between the current study and the Danish study we realize that we made a statement that was easily misunderstood. We did not intend to say that the two studies contradicted each other, but rather that they investigated different research question. The Danish study assessed predictors of post-disaster healthcare consumption (primary and specialized healthcare services pooled together) and showed that pre-disaster history of services consumption was a significant

predictor. In our study, we did not investigate post-traumatic distress or pre-disaster healthcare consumption as a predictor of post-disaster healthcare consumption. In our study, we investigated ratios of post- versus pre-disaster healthcare consumption. We found that a large proportion of the specialized mental healthcare services accessed following the attack were accessed by individuals that had not previously accessed healthcare. Thus, the two studies do not contradict each other, but rather answer different questions.

It could well be that natural disasters and terrorist attacks produce different healthcare needs in parents, as suggested by reviewer #1. However, comparisons of the current study and the Danish study are unable to inform such a hypothesis.

In order to avoid confusion on this matter, and to highlight how differences in disaster characteristics may produce different patterns of post-disaster healthcare consumption, the paragraph has been rewritten and now reads: “In the aftermath of disaster, an increase in healthcare consumption may result from both new patients entering the healthcare system, as well as an increase in frequency of service consumption among those already in the services (45). In our study, most participants were found to utilize primary healthcare services both before and after the terrorist attack. Thus, the increased primary healthcare service consumption stemmed largely from an increase in frequency of healthcare consumption in individuals that were known to the services. In contrast, the specialized mental healthcare services faced an influx of patients that were largely new to the services. Potential differences in parents’ post-disaster healthcare consumption according to disaster characteristics, e.g. magnitude, duration and potential for damage, whether it is a shared or a non-shared trauma of parent and child, whether it is a natural or a man-made disaster, as well as the levels of post-disaster psychosocial support are still largely to be investigated.”

Comment 8. In relation to the last point above, and the lack of literature in this area, I would suggest that the authors are even more tentative in their statement (p. 14) that similar patterns of healthcare need may ensue after other traumatic exposures of parents. Also, in relation to this statement, it was unclear whether the authors were talking about potentially traumatic events that directly impact only children.

Response: We thank reviewer #1 for this comment. We have carefully read through the manuscript in order to avoid unwarranted generalization of our findings. Furthermore, the sentence in the limitation section referred to by the reviewer (p.15) has been revised: “Whether similar patterns of healthcare needs arise in parents after other types of shared or non-shared traumatic exposures, such as when a child is struck by a natural disaster, traffic accident or serious illness, remains to be investigated.”

Reviewer #2

This manuscript provides important information about health service usage among parents of children exposed to a terror attack. As the authors point out, this severe event counted as a Criterion A trauma for parents as well as children, but indirect exposure to trauma in parents is often overlooked. The current manuscript provides a compelling case for the need to consider prolonged psychological distress following such an exposure in parents. I have the following minor comments.

Comment 1. There are many grammatical errors that need addressing throughout. Sometimes these result in errors in reporting (e.g., on page 13 – “only a majority of parents” should be a minority). This needs careful attention in any revision.

Response: We thank reviewer #2 for pointing out this critical mistake. In response to the comment, we have thoroughly reread the paper and had it proofread by a native English speaker.

Comment 2. In the methods, please can the authors provide brief summary of their original sample, versus simply referring to another publication, so that the extent to which was representative of the relevant parent population can easily be ascertained?

Response: In response to this comment, we have added information on the full parent sample. The paragraph now reads (p.7): “In the overall study, 299 mothers and 233 fathers (n=532 parents) participated in at least one of the three waves; 75.5% of the mothers (n=226) and 60.5% of the fathers (n=141) took part in Wave 3, on which this paper reports. The participants represented 251 distinct families and cared for a total of 263 survivors (54.6% of all Utøya survivors aged 13 to 33 years).”

Comment 3. The statistical treatment needs more explanation in order to be transparent for a non-expert reader, particularly the choice of regression model.

Response: In response to this comment and comments #5, #6 and #7 by reviewer #1, the statistics section has been rewritten (p9-10). Specifically, the following sentence has been added:
“Negative binomial hurdle regressions were chosen for rate predictions, as this method is suitable for overdispersed count data exhibiting excess zeros (39).”

To what extent do statistical analyses take account of time sensitive information presented in detail in the second figure, versus simply combine data over periods specified?
The statistical analyses combine data over the time periods specified (pre-disaster, early aftermath and delayed aftermath). The “observational period” has been explained in more detail in response to comment #5 by reviewer #1. (We are uncertain what the reviewer means by the term “time sensitive information”).

Comment: Clarifying the link between figures and key statistical analyses would be helpful - as far as I can tell analyses are represented by Figure 2, with other data being descriptive, but I wasn't certain.

Response: There may have been some problems with the figure numbering in the distributed material. Figure 2 is purely descriptive figure (line and pie chart), whereas Figure 3 reports the results of the regression analyses. We have received the material distributed to the reviewers, and see that the figures were not clearly numbered. Furthermore, legends appeared on a different page to the figure. In a publication the figures and labels will appear alongside, but we hope that the following information is helpful in review of the revised paper.

Descriptive information is provided in the following figures:
Figure 1: violin diagrams.
Figure 2: line and pie charts. Supplementing Figure 2 is the Supplementary table 3.
Figure 4: histograms. Supplementing Figure 4 is the Supplementary table 5.

Output of analyses is provided in the following figure:
Figure 3: Rate ratios with 95% confidence intervals. Supplementing Figure 3 is the Supplementary table 4.

All figures and Supplementary tables are cross-referenced through their labels (e.g. the legend to Supplementary table 4 includes the information: “Supplement to Figure 3.”).

In response to this comment, legends of the supplementary tables have been revised for clarity. (Furthermore, the Editor may choose to include references to the supplementary material in the text in the results section, rather than in the legends, at his/her discretion.)

Several specific points also need clarification. What were the results of analyses based on only in person consultations (referred to as being in supplementary material, but there are no data that duplicate Figure 2 and results are not clear).
Supplementing Figure 2 is the Supplementary table 3. This supplementary table contains both the results of the “all services” and “in person only” analyses. The legend of Figure 2 has been revised in order to highlight this: “Figure 2 a-b: Healthcare service consumption across time, presented as rates of services utilized (line chart) and proportions of mothers and fathers provided for within each six-month period (pie chart). Corresponding numeric values and values for in person consultations only are available in Supplementary table 3.”

Comment: What does it mean that ‘improved quality of the NPR registry during the pre-disaster period was observed’ and what did associated sensitivity analyses show?

“Improved quality of the NPR registry” refers to improved reporting practices by the healthcare providers across the study period. The improved reporting practices is reflected in lower levels of missing patient identification number (ID) in the registry across the study period, as shown in Supplementary table 2.

Response: In response to this comment, the description of data quality (p8) has been revised. It currently reads: “Quality of data. Claims to the national insurance scheme (HELFO) are submitted electronically. All claims with missing patient IDs are automatically rejected and returned to the healthcare provider for resubmission. Thus, the HELFO database contains no data with missing patient ID. In contrast, NPR does not reject incomplete information, and consequently contains a small number of entries with missing patient ID (Supplementary table 2).”

In the statistics section (p10), the sensitivity analyses are currently explained by the following revised paragraph: “Across the time period investigated, improved quality of reporting practices to NPR was observed (fewer entries were recorded with missing patient IDs, Supplementary table 2). The improved quality of reporting practices was most evident between 2009 and 2010. In the final year pre-disaster, levels of missing patient IDs were not substantially different to post-disaster levels. As incomplete entries in NPR may lead to underestimation of healthcare consumption, sensitivity analyses were performed by repeating all analyses that included the three-year pre-disaster NPR data, with NPR data from the final year pre-disaster only.”

In the results section (p12), the following revised sentence reports the results of the sensitivity analyses: “Sensitivity analyses addressing the improved reporting practices to NPR across the study period consistently returned conclusions that were not appreciably different to the findings presented in this paper, and are not shown.”

Comment 4. Tables/figures need improvement in order to be accessible. Figure 2, which seems to provide the key information, is difficult to process and I wonder whether some of this material could simply have been tabulated. Overall, figures seemed overly complex, need further annotation in order to be completely clear. It currently isn't clear what distinct, essential information each figure provides. The text refers to colours in violin figures that were not represented in the manuscript. Supplementary tables are even less accessible, and not all are referred to in the text so it wasn't clear what information was intended to be extracted from each.

Response: We thank reviewer #2 for these important comments. We have added the following to the statistics (p9): “Descriptive statistics are presented graphically. The distribution of overall frequencies by which healthcare services were accessed before and after the terrorist attack are presented as split violin diagrams (37).

Rates of healthcare service consumption across the study period and the proportions of individuals accessing healthcare services semiannually are presented in a second figure, by line and pie charts. As is often found in data on healthcare consumption (38), our data was overdispersed (variance greater than the mean value) and exhibited excess zeros (individuals with no occurrences). The corresponding numerical values are tabulated in the supplementary material that mirrors the graphics included in the paper." Furthermore, all figure and table legends have been revised. Reference to the colors of the figures has been removed from the text (however we think that the figures are easier to interpret with colors).

When the reviewer is referring to Figure 2, could it be that she is in fact referring to Figure 3? (Figure 2 is purely a descriptive figure consisting of line and pie charts. Figure 3 reports the results of the regression analyses). We understand that the figures were not clearly labeled in the material distributed to the reviewers and that the labels did not accompany the figures (on the same page). We believe that this may have made the link between the figure and the supplemental material difficult. For numbering of the figures in the review process, please refer to our response on comment #3 (explaining the numbering of the figures and the associated supplementary material).

Comment 5. In the discussion, gender differences might be further unpacked. In particular, more in depth consideration of whether this can be interpreted as due to a difference in actual rates of mental health problems or a difference in rates of treatment access would be helpful. It might also be useful if any brief reference to the potential financial burden of the increased healthcare usage could be made.

Response: We thank reviewer #2 for this input on the discussion. We have added the following to the discussion (p.13): "Increase in post-disaster primary healthcare consumption was most notable in female participants. Previous research has identified female survivors of trauma as more susceptible to developing post-traumatic stress disorder than men (40, 41). Thus, our data may in part reflect gender differences in post-disaster stress reactions. However, an alternative explanation is that distressed fathers may have been more reluctant than mothers to seek help for mental health complaints, as has been suggested by previous research (42, 43). Potential barriers to accessing post-disaster healthcare in men and women should be further addressed in future research."

Regarding the financial burden, we did not address this in this study. As the number of individuals directly impacted by the terrorist attack was small, the financial burden for the society was considered minor. Nevertheless, the financial burden for the individual parent may have been higher, e.g. as their capability of generating income may have been reduced. A discussion on financial burden is complex. We consider it to be beyond the scope of the paper.

Finally, we wish to thank both reviewers again for their valuable input on this manuscript.