PEER REVIEW HISTORY

BMJ Paediatrics Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

ARTICLE DETAILS

TITLE (PROVISIONAL)	Maltreatment, homicide, and access to specialized palliative care among children and young adults with a life-limiting condition: A nationwide population-based study
AUTHORS	Lin, Shih-Chun; Chang, Hsin-Yi; Huang, Mei Chih

VERSION 1 - REVIEW

REVIEWER NAME	***Po-Hsuan Lai***
REVIEWER AFFILIATION	E-Da Hospital
REVIEWER CONFLICT OF INTEREST	
DATE REVIEW RETURNED	19-Apr-2024

GENERAL COMMENTS	The introduction, Aim of the study, and Methods are well written. On page 7, I would recommend adding "Fraser's" in front of "International Classification of Diseases 10th revision" for clarity. Page 11: What other contributing influences would affect care burden and palliative care referrals, such as age, diagnosis, parental education, etc? Did the authors consider the influence of unnatural death may related to acute death, thus decreasing the palliative care rate? Another area for the authors to develop is the impact of culture and society on the response to palliative care use. Did the authors consider underestimation of maltreatment cases through using the sort of manner of death?

REVIEWER NAME	***Peter Flom***
REVIEWER AFFILIATION	Peter Flom Consulting
REVIEWER CONFLICT OF INTEREST	
DATE REVIEW RETURNED	10-Oct-2024

GENERAL COMMENTS	I confine my remarks to statistical aspects of this paper. These were quite simple, but appropriately so, so I vote to accept.

REVIEWER NAME	***Peter Sidebotham***
REVIEWER AFFILIATION	None disclosed
REVIEWER CONFLICT OF INTEREST	
DATE REVIEW RETURNED	16-Oct-2024

GENERAL COMMENTS	This is an interesting population-based study that attempts to address two important clinical issues: first, whether children and

	young adults with life-limiting conditions are more likely than those without to suffer maltreatment or unnatural death; and second, whether children and young adults with life-limiting conditions who die having experienced maltreatment or unnatural death are less likely to have received specialised palliative care than those who have not experienced maltreatment or unnatural death. The authors set out the background and rationale for their study clearly and succinctly. The hypotheses are expressed as 'children and young adults with LLCs had higher risks of maltreatment and unnatural death' and 'those with LLCs who reported maltreatment had less access to SPC'. These hypotheses are linked to two study goals. I have some reservations about both hypotheses: 1. While it is a reasonable hypothesis, justified by prior literature, that children and young adults with LLCs may have higher risks of maltreatment, the same does not hold for unnatural death. In their classification of unnatural deaths, the authors include both accidents and suicides, both of which typically involve a degree of independence on the part of the victim. Many children and young adults with LLC will be more dependent on their carers, and will often be less physically mobile. Thus, it is not unreasonable to hypothesise that they would have LOWER risks of accidental deaths and suicides. A more meaningful hypothesis would be that they have higher risks of maltreatment and homicide. If this were used as an hypothesis, the accompanying research goal and method would need to be amended to just include those deaths from homicide (15 and 61 respectively in the LLC and non-LLC groups). This would, in turn, influence the findings and conclusions. 2. While the second hypothesis is appropriate, the second study goal goes beyond this to examine whether those with LLCs who reported maltreatment OR died as an unnatural death received less SPC. This should be clarified in the expression of this hypothesis. As above, the inclusion of those who died of accidents or suicides similarly influences the findings and introduces a different issue. 3. The inclusion of unnatural deaths within the second hypothesis adds a further complication which potentially invalidates the hypothesis. By their very nature, homicides, suicides and accidental deaths are sudden events, which may result in death prior to or shortly after arrival at hospital. Specialist palliative care services are typically accessed for children whose health is deteriorating and are requiring additional support and intervention. This implies either that there is time between their admission to hospital and their death to initiate the palliative care services, or that they have longer-term needs requiring more intervention, and thus implying greater dependency and less independence. Thus, those with LLC who die of unnatural causes would be LESS likely to receive SPC. In my opinion, this second hypothesis would be better focused solely on those with recognised maltreatment prior to death, and the research goal and method adjusted accordingly. The methods are appropriately described in keeping with the above hypotheses, but will need amending if the hypotheses are adjusted as suggested above. Some further limitations are apparent from the methods: 1. The identification of maltreatment is dependent solely on hospital diagnoses of maltreatment between January 2016 and December 2017 as coded presumably by hospital staff. This presumably does not include children whose maltreatment was identified outside the

	hospital, for example through the police or social services. It also excludes any children with maltreatment identified before this time frame. 2. The identification of LLC is dependent on a hospital diagnosis and relevant coding during 2015. What about any children with LLCs diagnosed before or after this time frame? 3. I am somewhat unclear as to the data collection for phase 3. In phases one and two, data for LLC were collected during 2015 and data on maltreatment and deaths in 2016-17. In phase 3, data on deaths appear to have been collected between 2009 and 2017, a much longer time frame. What about data on LLC and on maltreatment for this phase? Were they also collected over the same time frame? This needs to be clarified. The results are presented clearly with an appropriate use of tables and text. For the proportion of maltreatment, a Chi-square calculation should be included (this is not a statistically significant difference). When reporting the numbers of unnatural deaths, it would be helpful to clarify that the majority of these were deaths from accidents. If the hypotheses and methods are amended to include only homicide deaths, the proportions are very different: LLC: 15/2,151 (0.6%) Non-LLC: 76/4,957 (1.5%) While this remains statistically significant it is very different from the 94% reduction in odds reported. If the second hypothesis is amended to compare those children with previous maltreatment who received SPC prior to death with those without previous maltreatment (and not including those who died of unnatural causes), the numbers would be very different. I recognise that this may prevent any meaningful analysis given that there were just 11 children and young adults with identified LLC and maltreatment. It may be that this hypothesis is set aside as not being testable with the current data. The discussion is appropriate given the results presented. It will however need amending in light of any changes to the hypotheses and methods. As it stands, the statement that those with underlying LLCs had a 94% decrease in the odds of unnatural death is correct, but somewhat meaningless given the arguments above regarding independence. Similarly to state that these children had an 87% rate reduction of SPC referrals may be correct but again is meaningless given the nature of these deaths. It is not correct to say that your results indicate that children and young adults with LLCs may be better protected and cared for than healthy people in the community – I don’t think your data are complete or sensitive enough to draw such a conclusion. In light of the above considerations, the strengths and limitations of the study need to be rethought, acknowledging further limitations. Minor issues: In the introduction, the authors refer to five types of child maltreatment (p. 5, line 41). I presume these are types recognised in Taiwan. There are other categorisations of child maltreatment used elsewhere. I would amend the statement to clarify this is the categorisation used in Taiwan.

	I am not clear why the code Y07, perpetrator of assault, maltreatment and neglect is included as definite maltreatment. Surely you are interested in children and young people as victims not perpetrators of maltreatment?

VERSION 1 – AUTHOR RESPONSE

Reviewer 1

The introduction, Aim of the study, and Methods are well written.

On page 7, I would recommend adding "Fraser's" in front of "International Classification of Diseases 10th revision" for clarity.

Response: We thank the reviewer for this comment and have added "Fraser's" in front of "International Classification of Diseases 10th revision" for clarity.

Page 11: What other contributing influences would affect care burden and palliative care referrals, such as age, diagnosis, parental education, etc?

Response: We appreciate the reviewer's comment. Due to the limited sample size, we were unable to control for these confounding factors, but we have included this as a limitation in the discussion section.

Did the authors consider the influence of unnatural death may related to acute death, thus decreasing the palliative care rate?

Response: We appreciate the reviewer for providing this comment. We agree that acute death decreases palliative care rates, so we changed our outcomes to focus on homicide deaths instead of other types of unnatural deaths.

Another area for the authors to develop is the impact of culture and society on the response to palliative care use.

Response: We appreciate the reviewer for providing this valuable comment. We have added an introduction and discussion regarding the barriers faced by healthcare professionals in initiating specialized palliative care (SPC) in Taiwan, as well as the disparities in access to SPC.

Did the authors consider underestimation of maltreatment cases through using the sort of manner of death?

Response: We appreciate the reviewer for bringing this to our attention and have included it as a limitation in the discussion section.

Reviewer 2

I confine my remarks to statistical aspects of this paper. These were quite simple, but appropriately so, so I vote to accept.

Response: Thank you.

Reviewer 3

This is an interesting population-based study that attempts to address two important clinical issues: first, whether children and young adults with life-limiting conditions are more likely than those without to suffer maltreatment or unnatural death; and second, whether children and young adults with life-limiting conditions who die having experienced maltreatment or unnatural death are less likely to have received specialised palliative care than those who have not experienced maltreatment or unnatural death.

The authors set out the background and rationale for their study clearly and succinctly. The hypotheses are expressed as ‘children and young adults with LLCs had higher risks of maltreatment and unnatural death’ and ‘those with LLCs who reported maltreatment had less access to SPC’. These hypotheses are linked to two study goals.

I have some reservations about both hypotheses:

1. While it is a reasonable hypothesis, justified by prior literature, that children and young adults with LLCs may have higher risks of maltreatment, the same does not hold for unnatural death. In their classification of unnatural deaths, the authors include both accidents and suicides, both of which typically involve a degree of independence on the part of the victim. Many children and young adults with LLC will be more dependent on their carers, and will often be less physically mobile. Thus, it is not unreasonable to hypothesise that they would have LOWER risks of accidental deaths and suicides. A more meaningful hypothesis would be that they have higher risks of maltreatment and homicide. If this were used as an hypothesis, the accompanying research goal and method would need to be amended to just include those deaths from homicide (15 and 61 respectively in the LLC and non-LLC groups). This would, in turn, influence the findings and conclusions.

Response: We appreciate the reviewer’s important feedback. We agree with the suggestion to revise our hypothesis regarding the statement that “children and young adults with LLCs faced greater risks

of maltreatment and homicide death." Consequently, we have updated the aim, methods, results, and conclusions to reflect this change.

2. While the second hypothesis is appropriate, the second study goal goes beyond this to examine whether those with LLCs who reported maltreatment OR died as an unnatural death received less SPC. This should be clarified in the expression of this hypothesis. As above, the inclusion of those who died of accidents or suicides similarly influences the findings and introduces a different issue.

Response: We appreciate the reviewer for this valuable comment. We agree with the reviewer and have changed the research question to determine "whether a record of maltreatment would affect the likelihood of receiving SPC referrals."

3. The inclusion of unnatural deaths within the second hypothesis adds a further complication which potentially invalidates the hypothesis. By their very nature, homicides, suicides and accidental deaths are sudden events, which may result in death prior to or shortly after arrival at hospital. Specialist palliative care services are typically accessed for children whose health is deteriorating and are requiring additional support and intervention. This implies either that there is time between their admission to hospital and their death to initiate the palliative care services, or that they have longer-term needs requiring more intervention, and thus implying greater dependency and less independence. Thus, those with LLC who die of unnatural causes would be LESS likely to receive SPC. In my opinion, this second hypothesis would be better focused solely on those with recognised maltreatment prior to death, and the research goal and method adjusted accordingly.

Response: We thank the reviewer for providing this valuable comment. We agree with the reviewer that those who experience acute death have fewer opportunities to receive SPC services. As a result, we have revised the research question to investigate "whether a history of maltreatment influences the likelihood of receiving SPC referrals." We have also adjusted the methods, results, and conclusions to reflect this change.

The methods are appropriately described in keeping with the above hypotheses, but will need amending if the hypotheses are adjusted as suggested above. Some further limitations are apparent from the methods:

1. The identification of maltreatment is dependent solely on hospital diagnoses of maltreatment between January 2016 and December 2017 as coded presumably by hospital staff. This presumably does not include children whose maltreatment was identified outside the hospital, for example through the police or social services. It also excludes any children with maltreatment identified before this time frame.

Response: We thank the reviewer for their helpful comment. We have added the following text to the discussion section, where we address this as a limitation:

“Second, identification of maltreatment was dependent solely on hospital diagnoses of maltreatment as coded presumably by hospital staff. This presumably does not include children and young adults whose maltreatment was identified outside the hospital, such as through police reports or social services, who did not seek formal medical care. Additionally, it did not account for cases identified outside of the research time frame.”

2. The identification of LLC is dependent on a hospital diagnosis and relevant coding during 2015. What about any children with LLCs diagnosed before or after this time frame?

Response: We appreciate the reviewer for bringing this to our attention. In Phase 1 of our study, we aimed to estimate the proportion of maltreatment among children and young adults with LLCs. We identified individuals who were documented as having an LLC in 2015 and who experienced maltreatment between 2016 and 2017. This approach ensures that we are considering individuals who still require medical care and are currently suffering from LLCs while facing maltreatment. We have also clarified this point in the methods section of Phase 1.

3. I am somewhat unclear as to the data collection for phase 3. In phases one and two, data for LLC were collected during 2015 and data on maltreatment and deaths in 2016-17. In phase 3, data on deaths appear to have been collected between 2009 and 2017, a much longer time frame. What about data on LLC and on maltreatment for this phase? Were they also collected over the same time frame? This needs to be clarified.

Response: Thank you for pointing out the unclear description. We have added clarification regarding the experience of maltreatment during the last year of life. It now states:

Phase 3

To compare SPC referral rates, we included all children and young adults aged 1-25 with a diagnosis of an LLC who died in Taiwan between January 2009 and December 2017. ...... We compared SPC referral rates for individuals with LLCs who had documented maltreatment one year prior to their death with those who had no documented maltreatment during the period from January 2008 to December 2017.

The results are presented clearly with an appropriate use of tables and text.

For the proportion of maltreatment, a Chi-square calculation should be included (this is not a statistically significant difference).

Response: We thank the reviewer for this recommendation and have included the results of the Chi-square calculation accordingly.

When reporting the numbers of unnatural deaths, it would be helpful to clarify that the majority of these were deaths from accidents.

Response: We thank the reviewer for picking this up. We have added the distribution of unnatural death types for patients with LLCs in both the results and discussions sections.

If the hypotheses and methods are amended to include only homicide deaths, the proportions are very different:

LLC: 15/2,151 (0.6%)

Non-LLC: 76/4,957 (1.5%)

While this remains statistically significant it is very different from the 94% reduction in odds reported.

Response: Thank you for bringing the unclear description to our attention. We have removed the following statement: “Those with LLC had a 94% decrease in the odds of unnatural death (OR, 0.06; 95% CI 0.05 to 0.07).” Instead, we have added this sentence: “Individuals with LLCs had a 68% decrease in the odds of dying from homicide (OR, 0.32; 95% CI 0.18 to 0.56)” in the results.

If the second hypothesis is amended to compare those children with previous maltreatment who received SPC prior to death with those without previous maltreatment (and not including those who died of unnatural causes), the numbers would be very different. I recognise that this may prevent any meaningful analysis given that there were just 11 children and young adults with identified LLC and maltreatment. It may be that this hypothesis is set aside as not being testable with the current data.

Response: We appreciate the reviewer highlighting this issue. We have re-evaluated our analysis and made the necessary revisions to the text:

“Patients with a maltreatment code were accounted for 1.1% (n = 14) of deaths (aged 1-25). 14.3% (2 out of 14) of children and young adults with a maltreatment code in the last year of life had received SPC. There was no significant difference in referrals to SPC at least three days before death between those with and without a maltreatment code (OR 0.52; 95% CI 0.12 to 2.33, p=0.39).”

The discussion is appropriate given the results presented. It will however need amending in light of any changes to the hypotheses and methods. As it stands, the statement that those with underlying LLCs had a 94% decrease in the odds of unnatural death is correct, but somewhat meaningless given the arguments above regarding independence.

Response: We have revised the discussion based on our amended hypothesis and results.

Similarly to state that these children had an 87% rate reduction of SPC referrals may be correct but again is meaningless given the nature of these deaths.

Response: We have revised the discussion to align with our amended hypothesis and results.

It is not correct to say that your results indicate that children and young adults with LLCs may be better protected and cared for than healthy people in the community – I don't think your data are complete or sensitive enough to draw such a conclusion.

Response: We thank the reviewer for bringing this to our attention and have removed the sentence from the discussion section.

In light of the above considerations, the strengths and limitations of the study need to be rethought, acknowledging further limitations.

Response: We thank the reviewer for this recommendation. We have expanded the discussions section, particularly focusing on a more in-depth discussion of the strengths and limitations of this study.

Minor issues:

In the introduction, the authors refer to five types of child maltreatment (p. 5, line 41). I presume these are types recognised in Taiwan. There are other categorisations of child maltreatment used elsewhere. I would amend the statement to clarify this is the categorisation used in Taiwan.

Response: We thank the reviewer for this comment. We have slightly altered the wording of the sentence. It now states:

"There are five common types of violence against children in Taiwan: violence exposure; physical abuse; emotional or psychological abuse; physical, emotional, or psychological neglect; and sexual abuse."

I am not clear why the code Y07, perpetrator of assault, maltreatment and neglect is included as definite maltreatment. Surely you are interested in children and young people as victims not perpetrators of maltreatment?

Response: We are grateful to the reviewer for identifying this issue, and we have deleted the code Y07 to ensure the accuracy of the results.