

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)	Access to paediatric palliative care in children and adolescents with complex chronic conditions: a retrospective hospital-based study in Brussels, Belgium.
AUTHORS	Friedel, Marie; Gilson, Aurélie; Bouckenaere, Dominique; Brichard, Bénédicte; Fonteyne, Christine; Wojcik, Thomas; De Clercq, Etienne; Guillet, Alain; Mahboub, Alaa; Lahaye, Magali; Aujoulat, Isabelle

VERSION 1 – REVIEW

REVIEWER	Reviewer name: Stuart Jarvis Institution and Country: University of York, UK Competing interests: None
REVIEW RETURNED	25-Jul-2019

GENERAL COMMENTS	<p>This is an interesting study in what appears (from the background information supplied by the authors) to be an under-researched area. I think it is useful in summarising the situation in the Brussels region, but there are limitations to the study which should be discussed in more detail as should the implications and importance of the study. Substantive issues follow, with minor suggestions for modifications to the manuscript to aid clarity following.</p> <p>Substantive issues:</p> <ul style="list-style-type: none"> - The authors present tables showing how hospital and PLT populations split between genders, age groups and conditions and how deaths and hospital admissions vary across the two groups. However, this is barely picked up in the results or discussion sections - what do these figures tell us and what are the implications? Are the distributions between groups significantly different (add 95% CI or chi-squared test for example?). The discussion should explain why any differences are important - are there particular groups that PLT fails to reach? - Missing data should be discussed. In particular, there are very many unknown conditions (26%) for PLT. This may make it impossible to tell whether the hospital and PLT groups differ (which is ok, if the data are simply not there, but should be acknowledged). Why are so many unknown? - I am not familiar with the context in Brussels, but on reading I wondered whether some patients attending Brussels hospitals but resident elsewhere may be referred to PLTs elsewhere (and also whether some patients at hospitals elsewhere may be referred to Brussels PLTs). These could explain apparently low PLT referral rates. Can the authors look at this at all? Either from the data or from knowledge of the healthcare systems - i.e. are there other regional PLTs that many patients may have gone to instead? It may just be a case of explaining the health system to those unfamiliar with it. Something for the discussion. The bottom right box of figure 2 suggests that patients from outside the area are sometimes referred to Brussels PLTs? Or that patients who have not had hospital
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	<p>admissions are referred to Brussels PLTs?</p> <ul style="list-style-type: none"> - What does this study add compared to reference 10? (For discussion and also introduction, for introduction to explain why do this study) - Finally, what are the implications? Is low referral to a PLT a bad thing that should be addressed? Or do most of the patients not need a PLT? Why do these results matter? <p>Minor issues/suggested wording revisions:</p> <ul style="list-style-type: none"> - Page 2 Line 22: I would use "Trusted Third Party" (i.e. capital T for Trusted) - Page 2 lines 26-29 appear to contradict each other - first says 572 referred to PLT, then only 384 referred to PLT. Just need to explain difference between the two figures (must refer to different things, but it's not clear) - Page 2 Lines 33: PLTS -> PLTs? - Page 3 Line 12: Reference 4 is for a study in England, not whole UK as implied. There is also the more recent ChiSP study (Scotland) if other UK results are considered relevant (disclosure: I am an author on that study). - Page 3 line 15: Studies relying on death registrations may miss a lot of individuals (I'm author on a paper https://doi.org/10.1177%2F0269216317728432) which is a good reason for instead taking your approach of using routine health data - Introduction, in general: It would be useful to explain the distinction (if any) between CCC and life limiting conditions - Page 4 Patient Selection: Do the two PLTs based in Brussels cover the 8 hospitals (i.e. would it be expected that patients at the 8 hospitals should be referred to these PLTs?) - Page 5 lines 14-15: were all the steering committee authors on the paper? I can see the relevance of mentioning discussions with an external body, but if the external body was only made up of the authors it seems less relevant - the authors on a paper normally discuss the results! - Page 5 lines 45-46: Were not referred to the two PLTs included in the study, but could have been referred elsewhere? - Page 6 line 7: clarify that you are referring to children admitted to hospital - Page 7 line 9: why are deaths unknown for those referred to PLTs aged 0-11 months? - Page 7 lines 16: "1-82 days" and "1-131 days" - should these be times, not days? They seem to be numbers of admissions not length of stay - Page 7 line 18: "Mean length of stay" -> "Length of stay"? As the numbers presented are median, range and mean. - Page 7 lines 30-33: Circular argument - non referral to PLTs explained by few being referred to PLTs. Just needs rewording. - Page 7 lines 35-37: What was not registered in hospital databases? The deaths? Or those children at all? - Page 7 line 58 - "accurate" -> "precise"? - Figure 2: Even though final size appears to be larger, text should perhaps be bigger (hard to read at least when presented in current size)
REVIEWER	<p>Reviewer name: Dr Timothy Warlow</p> <p>Institution and Country: All Wales Paediatric Palliative Care Network, Wales, UK</p> <p>Competing interests: None</p>
REVIEW RETURNED	30-Jul-2019

GENERAL COMMENTS	<p>This is a clear and focussed study. The methodology is clearly explained and achieves the initial aims of the study. Data is displayed clearly and I especially like the table displaying how data was aggregated. Early on in the study the reader suspects that your population of children with CCCs is in fact very broad. Many of these patients are unlikely to meet criteria for most tertiary paediatric palliative care teams either by prognosis (the surprise question) or disease category as used by Fraser et al criteria or the Hain directory. This may explain a large proportion of the low referral rate. However, you openly acknowledge these limitations and do not overstate the link between your CCC population and referrals to palliative care. I would agree that it is likely that more patients would benefit from palliative care services based on your data and I think your study provides some preliminary evidence to support that.</p> <p>My only suggestion would be to use 'children and adolescents' rather than the 'CA' abbreviation through the study as this flows better for the reader in my opinion.</p> <p>Wishing you every success in children's palliative care in Belgium.</p>
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VERSION 1 – AUTHOR RESPONSE

Reviewer 1 comments

1. Comments : This is an interesting study in what appears (from the background information supplied by the authors) to be an under-researched area. I think it is useful in summarising the situation in the Brussels region, but there are limitations to the study which should be discussed in more detail as should the implications and importance of the study. Substantive issues follow, with minor suggestions for modifications to the manuscript to aid clarity following.

Response: Thank you for this comment.

2. Substantive issues : The authors present tables showing how hospital and PLT populations split between genders, age groups and conditions and how deaths and hospital admissions vary across the two groups. However, this is barely picked up in the results or discussion sections - what do these figures tell us and what are the implications? Are the distributions between groups significantly different (add 95% CI or chi-squared test for example?). The discussion should explain why any differences are important - are there particular groups that PLT fails to reach?

Response: We developed those differences in the discussion section of the revised version.

Please find herunder the p values for gender, age and diseases categories :

P-values (chi-squared test):

Gender: 0.4523

Age: 0.02474

Diseases: < 0.001

Deaths: unknown => impossible to calculate

P-values (median test):

Frequency: < 0.001

Length of stay: <0.001

We added in the discussion section that children between 0-1 year are currently under referred to paediatric liaison teams, because neonatal palliative care is unsufficiently recognized in Belgium.

3. Missing data should be discussed. In particular, there are very many unknown conditions (26%) for PLT. This may make it impossible to tell whether the hospital and PLT groups differ (which is ok, if the data are simply not there, but should be acknowledged). Why are so many unknown?

Response: The high proportion (26%) of unknown conditions in the paediatric liaison groups can be explained by the following facts : Registration of data started only in 2010, year of the accreditation of the PLTs. In fact, the registration system at the beginning was imperfect, data on diagnosis was unsufficiently registered, above all not following the ICD system. This lead to uncomplete data (principal diagnosis was unknown in 26% of the cases) which was, in our study, marked as unknown data.

4. I am not familiar with the context in Brussels, but on reading I wondered whether some patients attending Brussels hospitals but resident elsewhere may be referred to PLTs elsewhere (and also whether some patients at hospitals elsewhere may be referred to Brussels PLTs). These could explain apparently low PLT referral rates. Can the authors look at this at all? Either from the data or from knowledge of the healthcare systems - i.e. are there other regional PLTs that many patients may have gone to instead? It may just be a case of explaining the health system to those unfamiliar with it. Something for the discussion. The bottom right box of figure 2 suggests that patients from outside the area are sometimes referred to Brussels PLTs? Or that patients who have not had hospital admissions are referred to Brussels PLTs?

Response: We agree with this hypothesis. Indeed, some patients admitted to a hospital in Brussels could have been referred to paediatric liaison teams outside Brussels.

But, overall, as a previous study (Friedel et al. 2018) showed, a maximum of 720 children/adolescents are cared on an annual basis at a national level.

This means that if 572 children are referred over a 5 year period to the 2 paediatric liaison teams based in Brussels (a rough mean of 110 children/year), the other 4 PLTs (outside Brussels) are caring for maximum (720-110) 610 children, which remains very low with regard of the (22533 :5 years) 4500 children diagnosed with a CCC in hospitals based in Brussels on an annual basis.

5. What does this study add compared to reference 10? (For discussion and also introduction, for introduction to explain why do this study)

Response: The reference 10 (Friedel et al. 2018) only looked at the number and the profile of children referred to paediatric liaison teams at a national level in Belgium. We tried in this study to look at all the children facing a complex chronic condition who might need to be referred to a paediatric liaison team but who are not...

6. Finally, what are the implications? Is low referral to a PLT a bad thing that should be addressed? Or do most of the patients not need a PLT? Why do these results matter?

Response: We do think that children with CCC, which can mostly be considered as life limiting or life threatening conditions, could benefit from paediatric liaison teams, because PLTs in Belgium are not only providing palliative care but also offer liaison care for children who might have a long life expectancy. PLTs can provide a whole range of care, from a simple phone call, to organisation and coordination of home care services and also end of life and bereavement care if necessary. Those different « depth of care » are currently not formally differentiated by the teams themselves, making it difficult to clearly affirm that all the children identified in this study should be referred to a PLT. Additionally, it depends also on the local resources, the family resources, abilities, needs and wishes to decide if a child should be referred or not to a PLT.

7. Minor issues/ suggested wording revisions : Page 2 Line 22: I would use "Trusted Third Party" (i.e. capital T for Trusted)

Response: We agree, and therefore put a capital letter for 'trusted' in the revised version: "Trusted Third Party"

8. Page 2 lines 26-29 appear to contradict each other - first says 572 referred to PLT, then only 384 referred to PLT. Just need to explain difference between the two figures (must refer to different things, but it's not clear)

Response: We agree that this is confusing.

The original version stated : « Over 5 years (2010-2014), 22 721 children/adolescents aged 0-19 years were diagnosed with a CCC. Of this number, 22 533 were admitted to hospital and 572 were referred to a PLT. Of the 22 533 children/adolescents admitted to hospital, only 384 (1.7%) were referred to a PLT. » and for more clarity, was changed into :

« Over 5 years (2010-2014) in the Brussels region, a total of 22 721 children/adolescents aged 0-19 years were diagnosed with a CCC. Of this number, 22 533 were identified through hospital registries and 572 through PLT registries. By comparing the registries, we found that of the 22 533 children/adolescents admitted to hospital, only 384 (1.7%) were also referred to a PLT.

9. Page 2 Lines 33: PLTS -> PLTs?

Response: Indeed, we harmonised in the revised version the abbreviation to PLTs every time it appears.

10. Page 3 Line 12: Reference 4 is for a study in England, not whole UK as implied. There is also the more recent ChiSP study (Scotland) if other UK results are considered relevant (disclosure: I am an author on that study).

Response: Thank you, we will correct this information. In the revised version "United Kingdom" is changed in "England" and we added the reference of the ChiSP study in Scotland.

11. Page 3 line 15: Studies relying on death registrations may miss a lot of individuals (I'm author on a paper <https://doi.org/10.1177%2F0269216317728432>) which is a good reason for instead taking your approach of using routine health data

Response: Thank you, we will add this reference to affirm that studies relying on death registrations might miss a lot of individuals and that it is therefore better to use routine health data.

12. Introduction, in general: It would be useful to explain the distinction (if any) between CCC and life limiting conditions

Response: We have added those definitions in the introduction in order to clarify the understanding of those terms.

13. Page 4 Patient Selection: Do the two PLTs based in Brussels cover the 8 hospitals (i.e. would it be expected that patients at the 8 hospitals should be referred to these PLTs?)

Response: Yes, this would be expected.

14. Page 5 lines 14-15: were all the steering committee authors on the paper? I can see the relevance of mentioning discussions with an external body, but if the external body was only made up of the authors it seems less relevant - the authors on a paper normally discuss the results!

Response: Yes, all the steering committee were mentioned as co-authors, because they discussed the design and the results of the study.

Additionnaly, as mentioned in the acknowledgment section, we requested the advice to 3 external persons regarding the results obtained : prof. Stéphane Moniotte at UCLouvain, Dr Lorna Fraser (University of York), Dr Richard Hain (Wales)

15. Page 5 lines 45-46: Were not referred to the two PLTs included in the study, but could have been referred elsewhere?

Response: Please see our response to comment 4.

16. Page 6 line 7: clarify that you are referring to children admitted to hospital

Response: We have clarified this in the revised version.

17. Page 7 line 9: why are deaths unknown for those referred to PLTs aged 0-11 months?
Because the data is missing.

18. Page 7 lines 16: "1-82 days" and "1-131 days" - should these be times, not days? They seem to be numbers of admissions not length of stay

Response: Indeed, it is an error which was corrected.

19. Page 7 line 18: "Mean length of stay" -> "Length of stay"? As the numbers presented are median, range and mean.

Response: Yes, length of stay. The word « mean » before « length of stay » was deleted in the revised version.

20. Page 7 lines 30-33: Circular argument - non referral to PLTs explained by few being referred to PLTs. Just needs rewording.

Response: We changed the wording in the revised version.

21. Page 7 lines 35-37: What was not registered in hospital databases? The deaths? Or those children at all?

Response: Yes, a lot of missing data was observed regarding the deaths in the hospital databases.

22. Page 7 line 58 - "accurate" -> "precise"?

Response: We changed the word 'accurate' into 'precise'.

23. Figure 2: Even though final size appears to be larger, text should perhaps be bigger (hard to read at least when presented in current size)

Response: We enlarged the font size (10 to 11) for Figure 2 in the revised version to facilitate the reading.

Reviewer: 2 comments Author's response

1. Comments : Dear Authors,

This is a clear and focussed study. The methodology is clearly explained and achieves the initial aims of the study. Data is displayed clearly and I especially like the table displaying how data was aggregated. Early on in the study the reader suspects that your population of children with CCCs is in fact very broad. Many of these patients are unlikely to meet criteria for most tertiary paediatric palliative care teams either by prognosis (the surprise question) or disease category as used by Fraser et al criteria or the Hain directory. This may explain a large proportion of the low referral rate. However, you openly acknowledge these limitations and do not overstate the link between your CCC population and referrals to palliative care. I would agree that it is likely that more patients would benefit from palliative care services based on your data and I think your study provides some preliminary evidence to support that.

Response: Many thanks for your comments.

In fact, and that is the point, the criteria to start the follow up of a child by a paediatric liaison team are not restricted to palliative care, but include also liaison care. Liaison care is difficult to define. And so are the criteria: which children would benefit from a paediatric liaison team? Hard work is currently done by the 5 official accredited paediatric liaison teams to better define the type of children cared for, the referral criteria, and to better harmonise data registration.

at My only suggestion would be to use 'children and adolescents' rather than the 'CA' abbreviation through the study as this flows better for the reader in my opinion.

Response: We agree, in the revised version we replaced the abbreviation "CA" by "children and adolescents".

3. Wishing you every success in children's palliative care in Belgium.

Response: Thank you very much.