PEER REVIEW HISTORY

BMJ Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form (**http://bmjopen.bmj.com/site/about/resources/checklist.pdf)** and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

ARTICLE DETAILS

TITLE (PROVISIONAL)	Protocol for a scoping review about ethics in transition programs for adolescents and young adults with neurodisabilities
AUTHORS	Bogossian, Aline; Gorter, Jan Willem; Racine, Eric

VERSION 1 – REVIEW

REVIEWER	William D. Graf, MD Connecticut Children's Medical Center, US
REVIEW RETURNED	26-Dec-2017

GENERAL COMMENTS	Bogossian, Gorter, and Racine provide an excellent protocol for assessing ethical principles in programs supporting transition from adolescence to emerging adulthood. The report is clear, well-written, and well-referenced. The protocol provides a framework for future studies.

REVIEWER	Jay Berry Boston Children's Hospital, Harvard Medical School, Boston, Massachusetts, U.S.A.
REVIEW RETURNED	02-Jan-2018

GENERAL COMMENTS	Thank you for the opportunity to review this study. I'm familiar with the value of publications on methods to describe future randomized trials. I have less experience assessing the value of such studies on methods for scoping reviews. I defer to the editors, who are in a much better position to judge this value. Obviously, the team for the current study has thoughtfully constructed rigorous methods that will be subsequently used for their review. However, I would have preferred the study team submit their full review for publication rather than submit the current paper solely on the methods. If they wish to proceed with the current paper as written, it might be best to consider a few revisions: (1) shorten / tighten the introduction; (2) consider a figure / table or some type of visual to present the conceptual model; (3) consider a discussion section that articulates how the methods for the scoping review contribute to the literature.

REVIEWER	Charlotte Hall University of Nottingham
REVIEW RETURNED	05-Mar-2018

GENERAL COMMENTS	The manuscript outline the protocol for conducting a scoping review. Suitable detail on methodology has been provided. Please spell out NDD the first time used on page 5

REVIEWER	Prof Marios Adamou

	UK University of Huddersfield
REVIEW RETURNED	05-Mar-2018

GENERAL COMMENTS	The authors make an apriori determination of five ethical principles which they expect the ethical challenges of transition to coalesce after their review. The authors do not discuss the philosophy ethics at all and just present "principles" that they developed without been clear as to how this selection was supported. It almost seems that they have a priori decided what the literature would show. The use of "neurodevelopmental disability" is too wide and includes mental health (not all of them incidentally such as ADHD) and physical disorders. Arguably any ethical issues in transition could be different between different disorders.

REVIEWER	Astrid Janssens University of Exeter Medical School, University of Exeter, UK
REVIEW RETURNED	13-Mar-2018

GENERAL COMMENTS	Dear authors,
	
	I was intrigued by the title of your manuscript and looking forward to read the full paper. Unfortunately, I found the paper confusing due to the use of multiple terms to identify the topic under study, the paper lacks focus and international relevance.
	
	ABSTRACT
	
	(Introduction): two sentences (2nd & 3rd) contain four different words to describe what you will be looking at: models, guidelines, practices, programs. I find this confusing. They are all different things, target different audiences (readers) and aim to do something different. This does not help the reader to capture the essence of the paper.
	
	After having referred to "models, guidelines, practices, programs" in the introduction, your methods states you will identify "problematic moral situations" as revealed in "transitional care" as well as "strategies and recommendations".
	
	    What is a problematic moral situation? And does this imply you will conduct primary research where you will identify these situations in transitional care?
	
	    After having stated in the introduction you would look at "models, guidelines, practices, programs"; your method section states you will look at "strategies and recommendations".
	
	Could the authors please check author guidelines: does BMJ Open allow references in the Abstract?
	
	INTRODUCTION

I do not find your introduction helpful. It does not make a clear statement what the problem is, and how this scoping review will help to set the scene as to what needs to be done (after you have identified gaps in the existing literature) in the field of "ethics in transition programs".

With regards to the introduction, I have two main questions / issues (and I will try to illustrate with examples after):

- Can you please specify what it is you will be looking for: could you define what ethics in transition covers? The introduction contains too many different terms, and it is unclear whether the authors refer to something different every time, or the same thing. Hence: 1) how do you define ethics (are they "ethical questions", "problematic moral situations") 2) where are you looking for ethics (in guidelines, models, practices, programs, strategies, recommendations)?
- Transition to adult services is not unique to Canada and a lot of research has been conducted around transition from child to adult services (for children with chronic physical and or mental health conditions); as you have chosen BMJ Open as your journal, I would expect the authors to situate the problem in a more international context.

Page 5

Line 8: Would the definition as described in Morris 2013 (1) cover what you mean with neurodevelopmental disabilities?

Line 27: "growing number of models and guidelines developed…" -> good opportunity to cite international literature.

Line 45: The authors state that "youth with neurodevelopmental disabilities [please insert abbreviation here] may experience…." Reference 8 is a *commentary*: could the authors use a stronger reference that uses findings from primary research to support this statement?

Line 26: authors talk about "models and guidelines". Line 48 authors talk about practices.

Ethics: Line 47: ethical questions; line 54: ethics principles page 6 Line 31: ethical tensions, line 40: ethical challenges; page 7 line 22: ethical concerns; line 31: ethical considerations

Page 6

Line 4-5: reference?

Line 11: could the authors give international examples?

Line 15 vs Line 20: authors move from young people's needs to "values and preferences" – what is the link or do the authors assume these to be identical?

Line 22: I will refer to this later in the method section: what the authors claim to do is not involvement IN the scoping review; they will share the findings after the scoping review is completed. Detailed description of an example of end user involvement in a (systematic) review can be found in (2)

Page 7:

Line 40: Can a scoping review provide evidence as to which practices might challenge or facilitate a successful transition? I believe this to be beyond the aims of a scoping review.

Methods and analysis

The method section does not satisfy my need for more clarity around the "ethics" in this scoping review.

Could the authors elaborate on the difference between research question 1 and 2?

I repeat my concern about scoping reviews and how to use the findings of a scoping review; according to the cited guidelines (Arksey & O'Malley), the outcome of the scoping review is to identify gaps in the literature and or identify the relevance of a full systematic review; using findings and conclusions from a scoping review to make policy and practice recommendations is premature.

I appreciate this is a scoping review, yet could the authors elaborate on the choice of terms included and other elements of the search:

- The authors suggest to screen titles (and not abstract) at Level 1 screening. I don't see the point of splitting the screening up in two levels. Also, I foresee issues in the choice of criteria used for level 1 screening: if you only use title, you might not be able to know whether the full paper is English / French (most paper included in the suggested databases will have the title translated in English); not sure you will be able to identify type of study from the title (qualitative or quantitative study). Hence, you will end up with a lot of "unclear" – why not combine title and abstract?
- I find a few key term missing in the search (example: transition)
- Could the authors elaborate on the choice of terms related to the "ethical concerns": why include autonomy, determination, empower and not responsibility, trust, values, …
- Can the authors explain how they will adjust their search for the different databases?
- Inclusion criterion b – does this imply you would exclude work including policy makers and or siblings of young people in transition?
- Inclusion criterion c – do you imply you will only include primary research (if not, this criterion could do with either

4

	further details or exclusion criterion to complete it)
	References
	• Please check reference 2 – authors missing + incorrect page (correct: 1291-4); I presume this is the same as reference 14
	• Please check how you cite in text (*Levac et al* and sometimes *Levac, Colquhoun*)
	• BMJ Open uses Vancouver with abbreviated journal titles
	International literature on transition
	Please check the MILESTONE study; see: http://www.milestone-transitionstudy.eu/
	Suggestions for reference to international literature on transition
	(3) Cochrane review evaluating effectiveness of interventions designed to improve the transition of care for adolescents from paediatric to adult health services;
	(4) International Delphi study to reach consensus on key elements and indicators of a successful transition
	1. Morris C, Janssens A, Tomlinson R, Williams J, Logan S. Towards a definition of neurodisability: a Delphi survey. Dev Med Child Neurol. 2013;55(12):1103-8. 2. Coon JT, Gwernan-Jones R, Moore D, Richardson M, Shotton C, Pritchard W, et al. End-user involvement in a systematic review of quantitative and qualitative research of non-pharmacological interventions for attention deficit hyperactivity disorder delivered in school settings: reflections on the impacts and challenges. Health Expect. 2016;19(5):1084-97. 3. Campbell F, Biggs K, Aldiss SK, O'Neill PM, Clowes M, McDonagh J, et al. Transition of care for adolescents from paediatric services to adult health services. Cochrane Database Syst Rev. 2016;4:Cd009794. 4. Suris JC, Akre C. Key elements for, and indicators of, a successful transition: an international Delphi study. J Adolesc Health. 2015;56(6):612-8.

VERSION 1 – AUTHOR RESPONSE

Reviewer comments and authors' response

Section	Comments	Author's response

General	Reviewer #1 Bogossian, Gorter, and Racine provide an excellent protocol for assessing ethical principles in programs supporting transition from adolescence to emerging adulthood. The report is clear, well-written, and well-referenced. The protocol provides a framework for future studies.	Thank you for these kind comments.
	Reviewer #2 Thank you for the opportunity to review this study. I'm familiar with the value of publications on methods to describe future randomized trials. I have less experience assessing the value of such studies on methods for scoping reviews. I defer to the editors, who are in a much better position to judge this value. Obviously, the team for the current study has thoughtfully constructed rigorous methods that will be subsequently used for their review. However, I would have preferred the study team submit their full review for publication rather than submit the current paper solely on the methods. If they wish to proceed with the current paper as written, it might be best to consider a few revisions: (1) shorten / tighten the introduction; (2) consider a figure / table or some type of visual to present the conceptual model; (3) consider a discussion section that articulates how the methods for the scoping review contribute to the literature.	(1) The introductory paragraph has now been streamlined, both in length as well as in the variation of concepts identified. (2) A figure has been developed to demonstrate the breadth of concepts. (3) We added a short statement indicating how scoping reviews add to the literature but remained succinct on this topic since this is an increasingly recognized type of literature review.
	Reviewer #3	Neurodevelopmental disabilities has

	The manuscript outlines the protocol for conducting a scoping review. Suitable detail on methodology has been provided.	been spelled out.
	Please spell out NDD the first time used on page 5	
	Reviewer #4 The authors make an apriori determination of five ethical principles, which they expect the ethical challenges of transition to coalesce after their review. The authors do not discuss the philosophy ethics at all and just present "principles" that they developed without been clear as to how this selection was supported. It almost seems that they have a priori decided what the literature would show.	Thank you for this feedback. Indeed we have struggled with this idea and recognize how this may narrow our aims to develop a comprehensive mapping of ethical situations. We have better explained our data extraction strategy.
	Reviewer #4 The use of "neurodevelopmental disability" is too wide and includes mental health (not all of them incidentally such as ADHD) and physical disorders. Arguably any ethical issues in transition could be different between different disorders.	In order to provide clarity on our use of this broad, non-categorical term, on page 4 we have inserted a definition of neurodisability developed through a Delphi study initiated by the Peninsula Cerebra Research Unit (PenCRU) (Morris et al, 2013). We found this a suitable option as this definition was developed by a large multidisciplinary group of professionals with the input of caregivers of people with ND for the purposes of research and organisation and evaluation of services.
Abstract	Reviewer #5 Two sentences (2nd & 3rd) contain four different words to describe what you will be looking at: models, guidelines, practices, programs. I find this confusing. They are all different things, target different audiences (readers) and aim to do something different. This does not help the reader to capture the essence of the paper	We have streamlined these sentences and now use the term "program" to specify the focus of our inquiry
	Reviewer #5 After having referred to "models, guidelines, practices, programs" in the introduction, your methods states you will identify "problematic moral situations" as	We realize that these terms introduce confusion and have proposed a typology of ethical concepts (Figure 1) to help indicate the spectrum of ethical issues we will be extracting in the

	revealed in "transitional care" as well as "strategies and recommendations". What is a problematic moral situation? And does this imply you will conduct primary research where you will identify these situations in transitional care? After having stated in the introduction you would look at "models, guidelines, practices, programs"; your method section states you will look at "strategies and recommendations".	review. We have also streamlined language about models, guidelines, practices throughout the paper.
	Reviewer #5 Could the authors please check author guidelines: does BMJ Open allow references in the Abstract?	We have removed references from the abstract.
Introduction	Reviewer #5 I do not find your introduction helpful. It does not make a clear statement what the problem is, and how this scoping review will help to set the scene as to what needs to be done (after you have identified gaps in the existing literature) in the field of "ethics in transition programs". With regards to the introduction, I have two main questions / issues (and I will try to illustrate with examples after): • Can you please specify what it is you will be looking for: could you define what ethics in transition covers? The introduction contains too many different terms, and it is unclear whether the authors refer to something different every time, or the same thing. Hence: 1) how do you define ethics (are they "ethical questions", "problematic moral situations") 2) where are you looking for ethics (in guidelines, models, practices, programs, strategies, recommendations)? Transition to adult services is not unique to Canada and a lot of research has been conducted around transition from child to adult services (for children with chronic	In streamlining the introduction, we hope that we have removed the ambiguity and confusion previously introduced by the range of terms. On Page 5, a figure has been introduced to illustrate the range of ethical issues we will be seeking to document in this review along with examples to provide clarity. The figure identifies a continuum of concepts spanning from ethically problematic situations that have occurred and have been documented to more abstract ethical issues that are anticipatory in nature, and that could be tied to unintended consequences of elements in the delivery of transition programs. We are aware of the international literature and how transition issues are indeed a global concern. Dr. Gorter has extensive experience on the topic of transition and a thorough understanding of the literature (North America, UK, Scandinavia, Australia) where most literature originates

	physical and or mental health conditions); as you have chosen BMJ Open as your journal, I would expect the authors to situate the problem in a more international context.	
	Reviewer #5 Page 5, Line 8: Would the definition as described in Morris 2013 (1) cover what you mean with neurodevelopmental disabilities?	We agree that including this definition will provide clarity to the types of populations/studies we will include in our sample. We have included this definition on Page 4.
	Reviewer #5 Page 5, Line 27: "growing number of models and guidelines developed…" -> good opportunity to cite international literature.	In tightening the introduction, this sentence has been removed. We recognize that the international literature is quite extensive and expect to capture insights from that work in our scoping review
	Reviewer #5 Page 5, Line 45: The authors state that "youth with neurodevelopmental disabilities [please insert abbreviation here] may experience…." Reference 8 is a commentary: could the authors use a stronger reference that uses findings from primary research to support this statement?	In tightening the introduction, this sentence has been removed. Unfortunately the cited review is one of the only sources of information on ethics in transition programs. We hope that the proposed review will help identify primary sources where ethical issues have been reported but may not have been explicitly identified as such.
	Reviewer #5 Page 5, Line 26: authors talk about "models and guidelines". Line 48 authors talk about practices.	We have streamlined the language in the paper, maintaining the term "program".
	Reviewer #5 Page 5, Line 47: ethical questions; line 54: ethics principles page 6 Line 31: ethical tensions, line 40: ethical challenges; page 7 line 22: ethical concerns; line 31: ethical considerations	In tightening the introduction, these terms have been streamlined.
	Reviewer #5 Page 6, Line 4-5 reference?	We could not identify the relevant concern.

	Reviewer #5 Page 6, Line 11 - could authors provide international examples	We could not identify the relevant concern.
	Reviewer #5 Line 15 vs Line 20: authors move from young people's needs to "values and preferences" – what is the link or do the authors assume these to be identical?	In tightening the introduction, these terms have been streamlined.
	Reviewer #5 Page 6, Line 22: I will refer to this later in the method section: what the authors claim to do is not involvement IN the scoping review; they will share the findings after the scoping review is completed. Detailed description of an example of end user involvement in a (systematic) review can be found in (2)	Thank you for this feedback and for providing an example of end user involvement. The method of review we follow proposes to engage stakeholders once preliminary findings have been culled from the literature. While we have not involved stakeholders in the conception of the scoping review, we will engage stakeholders through participatory methods to discuss the meanings, gaps and opportunities of the preliminary findings, as well as seek counsel regarding dissemination and knowledge sharing. Co-authors are aware of the principles of stakeholder engagement and involvement As Director of the CanChild Centre for Childhood Disability Research, Dr. Gorter has extensive experience with conducting research utilizing principles of engagement and involvement https://www.canchild.ca/en/research-in-practice/knowledge-translation-exchange
	Reviewer #5 Page 7: Line 40: Can a scoping review provide evidence as to which practices might challenge or facilitate a successful transition? I believe this to be beyond the aims of a scoping review.	We will be drawing from the literature to synthesize information from published studies about practices that others have reported as challenging or facilitating successful transition.
Methods and analysis	Reviewer #5 The method section does not satisfy my	We have infused our signposts in the introduction of this paper including a figure that identifies the degrees of

	need for more clarity around the "ethics" in this scoping review.	ethical issues we will seek in the literature (spanning from concrete and realized problematic situations to abstract and anticipated ethical issues).
	Reviewer #5

Could the authors elaborate on the difference between research question 1 and 2? | We are making a distinction between ethical issues that may be embedded in the *vision* of transition programs (ie: to ensure youth can manage their own healthcare or youth can advocate for themselves) from ethical issues that may be embedded in the service delivery guidelines (ie: attention may be paid to ensure that the *transfer* of particular pediatric patients to adult healthcare settings). |
| | Reviewer #5

I repeat my concern about scoping reviews and how to use the findings of a scoping review; according to the cited guidelines (Arksey & O'Malley), the outcome of the scoping review is to identify gaps in the literature and or identify the relevance of a full systematic review; using findings and conclusions from a scoping review to make policy and practice recommendations is premature. | We appreciate this feedback. We have adjusted the wording in RQ 5 to read: "What key policy and practice recommendations can be ~~developed~~ *identified* from this knowledge synthesis?" to reflect our intention to identify and document key policy and practice recommendations from existing empirical work. In our concluding remarks, we propose to produce a "preliminary, literature-based, repository of policy and practice recommendations". |
| | I appreciate this is a scoping review, yet could the authors elaborate on the choice of terms included

and other elements of the search:

• The authors suggest to screen titles (and not abstract) at Level 1 screening. I don't see the point of splitting the screening up in two levels. Also, I foresee issues in the choice of criteria used for level 1 screening: if you only use title, you might not be able to know whether the full paper is English / French (most paper included in the suggested databases will have the title translated in English); not sure you will be able to identify type of study from the title

(qualitative or quantitative study). Hence, you will end up with a lot of "unclear" – why | Based on these helpful recommendations, we have infused changes in the methods section of the paper. Specifically, we have introduced the following changes:

- First level screening will include screening of title and abstract.
- The criteria to screen for quantitative or qualitative studies has been removed.
- The keywords and index terms identified in the paper refer to PubMed based MeSH terms. These keyword and index terms were adapted to the fit the terms used on the other different databases.
- We will include full search terms in addenda.
- We have expanded our definition of key stakeholders to include family |

11

	not combine title and abstract?	member, program managers and policy makers.
	• I find a few key term missing in the search (example: transition)	
	• Could the authors elaborate on the choice of terms related to the "ethical concerns": why include autonomy, determination, empower and not responsibility, trust, values, …	
	• Can the authors explain how they will adjust their search for the different databases?	
	• Inclusion criterion b – does this imply you would exclude work including policy makers and or siblings of young people in transition?	
	• Inclusion criterion c – do you imply you will only include primary research (if not, this criterion could do with either further details or exclusion criterion to complete it)	
References	Reviewer #5 • Please check reference 2 – authors missing + incorrect page (correct: 1291-4); I presume this is the same as reference 14 • Please check how you cite in text (Levac et al and sometimes Levac, Colquhoun) • BMJ Open uses Vancouver with abbreviated journal titles	We have cleaned up references and have standardized our in-text citations
International literature on transition	Reviewer #5 Please check the MILESTONE study; see: http://www.milestone-transitionstudy.eu/ Suggestions for reference to international literature on transition (3) Cochrane review evaluating effectiveness of interventions designed to improve the transition of care for adolescents from paediatric to adult health services;	At this time, we consider premature to cite a specific transition program since we aim to review them in the scoping review. In terms of international literature, we have cited both papers in order to reflect the international importance of transition programs.

	(4) International Delphi study to reach consensus on key elements and indicators of a successful transition	

VERSION 2 – REVIEW

REVIEWER	Astrid Janssens Institute of Health Research - Child Mental Health Group, University of Exeter Medical School, UK
REVIEW RETURNED	03-May-2018

GENERAL COMMENTS	Dear Authors, Thank you for your extensive review of this manuscript and your helpful responses to comments. I would like to congratulate you on the significant improvement of this version of your manuscript. This revised manuscript is so much easier to read, has a good flow, and streamlined use of language (terminology). The additional Figure is very helpful: it defines the framework (previously assumed) underlying your review / work and makes the paper more accessible. I would like to make a few more suggestions and will leave it to the Editor's discretion whether these comments and or suggestions require a response. ABSTRACT (pg2) You use the abbreviation ND once; either have the abbreviation between brackets first time (and use consistently throughout the abstract), or write in full. Introduction: the objective of the review is well described; line 15: the objective of this review is *to* identify… METHODS AND ANALYSIS Scoping review protocol design (pg 7-8; up until "stage 1"): this is rather lengthy; the many references in text (example line 11-16) hamper the flow. This section could be written up more succinctly which would benefit readability. Full stop missing, line 44 Pg 10: Stage 3 study selection Line 24-25: if a study is coded as unclear from the study title AND ABSTRACT, the study will be moved to the second level screening. -> the second level screening is not defined further in the protocol. Do the authors imply "study selection method" to be the second level screening? Stage 4: conceptual framework Very helpful additions to this section. Pg 11, line 3-4: can the scholarship title be removed from the sentence and replaced by a reference? Line 11: programS (should this be plural?) Line 29 + Line 34: Is research design not part of the study characteristics? Line 52: Full stop missing.

VERSION 2 – AUTHOR RESPONSE

Reviewer comments and authors' response

Section	Comments	Author's response
ABSTRACT (pg2)	You use the abbreviation ND once; either have the abbreviation between brackets first time (and use consistently throughout the abstract), or write in full.	Abbreviation ND has been replaced with "neurodisabilities". All other abbreviations have been removed: • AYA (replaced by "adolescents and young adults") • HCT (replaced by "health care transitions").
	Introduction: the objective of the review is well described; line 15: the objective of this review is *to* identify…	We have added the word "to" in the sentence "the objective of this review is "to" identify"
METHODS AND ANALYSIS	Scoping review protocol design (pg 7-8; up until "stage 1"): this is rather lengthy; the many references in text (example line 11-16) hamper the flow. This section could be written up more succinctly which would benefit readability.	We have streamlined the description of the scoping review protocol design, replacing references to specific authors with only in-text references and editing the text.
	Full stop missing, line 44	Full stop inserted
Pg 10: Stage 3 study selection	Line 24-25: if a study is coded as unclear from the study title AND ABSTRACT, the study will be moved to the second level screening. -> the second level screening is not defined further in the protocol. Do the authors imply "study selection method" to be the second level screening?	Thank you for identifying this confusion, we have modified the description for study selection and "unclear" studies.
Page 11, Line 3 - 4	can the scholarship title be removed from the sentence and replaced by a reference?	We have replaced the term "scholarship" with the term "literature". This framework was developed through a synthesis of studies about ethics in transitional care. The references for this literature can

		be found in the supplementary file.
	Line 11: programS (should this be plural?)	"s" added to the word program
Line 29 + Line 34	Is research design not part of the study characteristics?	We have modified the description on line 29 "We will also record additional information about aspects of the study design related to stakeholder involvement in the research design"
Page 11, Line 52	Full stop missing.	Full stop inserted