Author’s response to reviews

Title: Relational autonomy: what does it mean and how is it used in end-of-life care? A systematic review of argument-based ethics literature

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(You may find a Word document attached as 'Cover Letter - Revision' for a clearer format)

RELATIONAL AUTONOMY: WHAT DOES IT MEAN AND HOW IS IT USED IN END-OF-LIFE CARE? A SYSTEMATIC REVIEW OF ARGUMENT-BASED ETHICS LITERATURE

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Preliminary remark from the authors: Changes and additions in the text are all marked in yellow highlighting.

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Editor Comments (blue coloured) with a point-by-point response by the Authors:

Dear Mr Gómez-Vírseda,

Your manuscript "Relational autonomy: what does it mean and how is it used in end-of-life care? A systematic review of argument-based ethics literature" (METH-D-19-00131) has been assessed by our reviewers. They have raised a number of points which we believe would
improve the manuscript and may allow a revised version to be published in BMC Medical Ethics. Their reports, together with any other comments, are below (...). If you are able to fully address these points, we would encourage you to submit a revised manuscript to BMC Medical Ethics. (...) I look forward to receiving your revised manuscript and please do not hesitate to contact us if you have any questions.

Answer from the authors: Thank you for offering us the chance to revise our manuscript. We have carefully considered the comments given by both reviewers and amended the manuscript accordingly. Please find a detailed account of the amendments and our argumentation below.

This is a good paper and whilst it has been returned with a request for 'major revisions' this is probably an overstatement of what is required. The comments from Reviewer 1 are the most substantive but offer guidance on improving what is already a good paper.

Answer from the authors: Thank you for your appreciation and encouragement. We would like to thank the editor and both reviewers for their comments. These enabled us to increase the quality of our review.

ROSALIND MCDOUGALL (REVIEWER 1)

This systematic review focuses on an important topic and, as the authors highlight, an area of increasing scholarly interest. The search is rigorously designed, conducted and reported. Tables and figures are clear, concise and well-communicated. The discussion section is insightful and clearly-written.

Answer from the authors: Thank you for your appreciation.

There are three areas where revisions are necessary, in my view:

1. Explaining the synthesis process.

The paragraph on extraction and synthesis (p.8) needs rewriting to better explain the process for readers unfamiliar with the QUAGOL approach. What is meant by a 'conceptual scheme'? Are the 'conceptual-ethical questions' the same as the research questions? What does it mean to produce a 'comprehensive overall structure'? What does the structure aim to describe? What is meant by 'the most relevant' meanings? Was it the most relevant or the most common?
Answer from the authors: Thank you for the observation. In order to meet this comment, we have added some clarifications and amends to the manuscript.

Answering to each raised question by the reviewer in this comment:

- **A ‘conceptual scheme’ is an outline which provides the concepts that appear relevant to answer the research question.** In addition to a concise definition in the text, we have decided to attach an example of a conceptual scheme as supplementary data online (see Additional file 1).

- **We agree that it could be rather confusing to first speak of ‘conceptual-ethical questions’ and later of ‘research questions’, apparently implying that these were different from each other. In fact, the ‘conceptual-ethical questions’ are the ‘research questions.** We have gained consistency by always referring to them as ‘research questions’ in the text. Meeting this comment help to avoid misunderstandings, indeed.

- **The ‘global scheme’ is the synthesis of the fifty individual conceptual schemes.** Different concepts coming from different publications were presented together and interrelated in a synthetic way. It aims at providing a ‘comprehensive overall response’ to our research questions, as it has been now clarified in the manuscript.

- **When we highlighted ‘the most relevant’ meanings, foundations, or uses of relational autonomy, we refer to those which provide more insight into our research questions.** The quality rather than the quantity was taken into account. However, when a meaning, foundation or use was particularly common, it was also highlighted by providing the list of references.

After all the amends and clarifications, we now think that readers, even those unfamiliar with the QUAGOL approach, will be able to follow the process of extraction and synthesis.

p. 8: For extraction and synthesis, we used the Qualitative Analysis Guide of Leuven (QUAGOL) approach, which consists of five preparatory, sequential steps [24]. First, articles were read and reread, highlighting the relevant parts and the main arguments presented. Second, we developed a narrative summary of these highlighted parts of the articles. The aim was to draw strong lines of argumentation and identify where in the article the main concepts appear. Third, a conceptual scheme for each publication was created. A conceptual scheme is a synthetic frame where different concepts that appear relevant to answer the research questions are presented and interrelated with each other (an example of a conceptual scheme is provided as Additional file 1). Each conceptual scheme was appraised separately by two authors (CGV and CG) so that we could objectively and accurately characterise each included publication. Both appraisers discussed the resulting conceptual schemes until they agreed on their adequateness. Fourth, these individual conceptual schemes were considered as a whole to search for relationships that would
produce a comprehensive overall response to our research questions. Here, we aimed to focus on our research questions even if the individual article’s main concern was somewhat different from these issues. We built up a separate global scheme that integrated the most relevant meanings, foundations, and uses of the concept of relational autonomy. This scheme was iteratively evaluated and checked against previous QUAGOL steps in order to ensure that it was consistent. In the final and fifth step, we synthesised a description and report of these results to be presented in the Results section of this review.

In the paragraph starting 'A fourfold structure emerged…' (p.9), it is not clear if the authors are describing the content of the articles or using the content as a starting point for their own further theorising. For example, when it is stated that 'relational autonomy is theoretically conceptualised using the adjusted understanding from the first two sections', is it the authors doing the conceptualising?

Answer from the authors: The fourfold structure is based on the analysis of the content of the 50 articles. This particular structure is not reported as such in any individual publication but is the result of synthesizing and analysing the content of all the individual publications. A clarifying sentence has been added to the manuscript wherever this confusion may have occurred.

p. 9: As a result of the analysis and synthesis of the fifty individual articles, a fourfold structure was conceived by the authors (Fig. 2). […] In summary, we present our results in four main sections. First, we introduce a simplified interpretation of individual autonomy in mainstream bioethics, as derived from the analysis of the included publications. […]

2. Structure/presentation of results, particularly ‘individualistic autonomy'

Given the way in which the research questions were framed, I had expected the results to be organised in terms of ‘meanings, foundations and uses' of relational autonomy. It was not immediately clear why the authors were starting with 'the origins of an individualistic conception of autonomy', as these seemed far from the key findings in relation to the review's focal concept of relational autonomy. The role of this section in the authors' overall interpretation of this literature became clearer later in the discussion section, but needs to be better explained and contextualised. Reordering the results and/or renaming this theme/section 'simplified individualistic autonomy' may assist in this.

Answer from the authors: While the ‘meanings, foundations, and uses of relational autonomy’ were our guiding research questions, literally restricting our results to these three items would have led to a regrettable loss of nuances and complexity. We prefer to structure the results in four sections that not coincide exactly with the research questions. In our opinion, these sections offer
a clearer presentation of the notion of relational autonomy, while they satisfactorily answer the research questions.

We think that the first and second parts (a portrayal of individualistic autonomy and a critique of this individualistic interpretation) are necessary in order to better appreciate one of the key findings of this review. As it was highlighted in the results and conclusions, many publications only provide a development of the notion of relational autonomy in contrast to a certain view of individual autonomy. Therefore, it becomes necessary to synthesize this portrayal of individual autonomy, before presenting the concept of relational autonomy, theoretically and practically developed.

Yet, we take into account the reviewer’s concern and try to respond to it. To do so, we have added an introductory paragraph which clarifies the role of these two sections in the Results part. There, we justify why to start with a presentation of ‘a simplified individualistic autonomy’ -as it was suggested by the reviewer-. Nevertheless, we preferred not to use the adjective 'simplified' in the section’s title, as it may sound too pejorative.

p. 9: As a result of the analysis and synthesis of the fifty individual articles, a fourfold structure was conceived by the authors (Fig. 2). The first two sections present and then criticize a simplified interpretation of individualistic autonomy, against which relational autonomy is often developed. These two preliminary steps are necessary in order to better apprehend the last two sections, where relational autonomy is elaborated in theory and in practice. […]

3. Implications for healthcare decision-making

At times in the paper, the practical implications of the review seem overstated. For example, 'Key findings are highlighted and considered in depth with the aim of reaching a clearer path towards better decision-making in real-world healthcare situations' (p.23) and "Completing this analysis was important, because it illuminates a path forward toward better decision-making in healthcare, in general, and in end-of-life care, in particular" (p.27). While the review provides a useful synthesis of literature, the benefits seem primarily scholarly rather than practical. The claims throughout about practical implications need to be explained and justified, or moderated.

Answer from the authors: We agree with the appraisal of the reviewer: “While the review provides a useful synthesis of literature, the benefits seem primarily scholarly rather than practical”. Indeed, we reach similar conclusions when we discuss the operationalisation of relational autonomy: “Our review has revealed some distance between theoretical approaches to relational autonomy and its operationalisation in end-of-life practices. […]” (p. 26). In order to meet this comment, we have moderated some seemingly overstated sentences about the practical implications.
p. 23: [...] Main findings are highlighted and considered in depth, with the aim of reaching a clearer path towards better decision-making in healthcare.

p. 27: [...] Completing this analysis was important, because it may illuminate a path towards better decision-making in end-of-life care healthcare. Further analysis is required, however, to reach consensus on how to develop a structured and standardised decision-making process. [...] Other issues:

The use of lettering in both the results and conclusions sections of the abstract was confusing, particularly as the two sets did not align. I suggest removing the lettering from the conclusions.

Answer from the authors: In order to meet this comment, we have removed the lettering from both the results sections of the abstract and the conclusions of the manuscript. By doing so, we expect to have gained some more clarity.

In the background section, the conceptual connection between relational autonomy and shared decision-making needs to be explicit, as does the connection between relational autonomy and advance care planning. This would also help to clarify why investigating literature at the intersection of relational autonomy and end of life is important/useful; this is currently unclear in the background section.

Answer from the authors: We have amended the manuscript in accordance with the editor’s comment.

p. 4: [...] Especially for end-of-life ethics, the need for a relational turn in the understanding of autonomy has led to a growing number of publications on shared decision-making [10-13] and advance care planning [14-16]. Both of these practices in end-of-life care are unsatisfactorily conceptualised in the classical individualistic framework. Relational autonomy emerges here as an interesting concept for these trends in end-of-life decision-making. Empirical research about relational autonomy in this field highlighted potential changes in the doctor-patient relationship and in physicians’ responsibility towards patients and their families [17].

Throughout the paper, the authors use the term 'anthropology' in a different sense to the usage that I am familiar with. It would be helpful to define this term at first usage to alert the reader to the specific sense in which it will be used.

Answer from the authors: Thank you for the useful comment. In our article, the term ‘anthropology’ is used in a purely philosophical way, as the underlying view/comprehension of
the human being. We are aware of the use of the term ‘anthropology’ in other disciplines, such as cultural anthropology, theological anthropology, linguistic anthropology, etc. To avoid this confusion, we have tried to minimize the use of the term as much as possible. Whenever it was unavoidable, we have clarified its use as ‘philosophical anthropology’ and we have given a general definition at its first usage.

p. 10: Any interpretation of autonomy is unavoidably underpinned by a certain view of what a human being is, in other words: by a particular philosophical anthropology. The anthropology derived from our analysis could be described in terms of

Inclusion criteria specify "development of a relational approach". Was this applied requiring 'development' specifically, or was 'use of' a relational approach also sufficient to meet this criterion?

Answer from the authors: To meet this comment we have simplified the way this inclusion criterion is formulated. A meaningful use of the relational approach to autonomy was considered sufficient to meet this criterion. This use needed to be active, significant or somehow meaningful. We did not consider a merely passive use of relational autonomy.

p. 7: Inclusion criteria were the following: (1) significant use of a relational approach to the concept of autonomy, (2) application of […]

The exclusion of book chapters (p.7) needs some justification.

Answer from the authors: We decided to restrict the search only to journal articles so the review becomes completely systematic and reproducible. Using other databases suitable for book chapters (such as Google Scholar) would have made impossible a systematic search with Boolean methods. Moreover, fifty articles were indeed a significant number of publications in order to reach saturated enough information, while it remains a manageable amount of data.

The long lists of references in the results section make the text difficult to read meaningfully. Could this information be captured in another format (and included as a supplementary file) and the in-text citations limited to a few examples in each case?

Answer from the authors: We are aware that long lists of references make sometimes the text difficult to read. We have tried to answer the reviewer’s concern by shortening the list of references with a dash, whenever there was a sequence of consecutive numbers. Nonetheless, we keep the option for in-text citations for the sake of a more systematic report of the references.
Choosing some examples, as suggested, have the risk of arbitrariness. Systematic reporting is a key feature of systematic reviews.

What is meant by 'constructive analysis' on p.14? I'm not sure that this sentence is necessary.

Answer from the authors: As the sentence may be confusing and does not provide essential information, it has been removed from the manuscript. Thank you for the comment.

p. 15: We now present the conceptualisation of relational autonomy as described in the included publications.

On p. 14, I found the following sentence confusing: 'We identified some specific ethical publications while doing our analysis'. Do the authors mean ethical approaches/methodologies?

Answer from the authors: Indeed, it is a mistake. We actually mean ‘ethical approaches’ and so it has been amended in the text. Thank you for your comment.

p. 15: We identified some specific ethical approaches while doing our analysis.

LISA DIVE, PH.D. (REVIEWER 2):

The experience of this referee is in theoretical bioethics and analytic philosophy, therefore the comments made reflect this background. I am not formally trained in the methods of systematic reviews, but have some familiarity with the bioethical literature on autonomy, including relational autonomy.

This manuscript is a systematic review of "argument-based" ethics literature about the concept of relational autonomy in the context of end-of-life care. Overall, in this reviewer's opinion the paper is of good quality and addresses an important topic by providing a comprehensive analysis of relational autonomy, a central concept in end-of-life care. The method of undertaking a systematic review draws out with some clarity certain features of how relational autonomy is conceptualised and applied in this area of bioethics, which is a valuable contribution to the literature. The authors also have an emphasis on understanding the concept to enable its deployment to improve decision-making in end-of-life care situations, which is a positive feature of their approach.

Paper quality: The paper is clearly written and well structured.
Answer from the authors: Thank you for your appreciation.

Suggestion for improvement:

My main suggestion is that the authors clarify what is meant by "argument-based literature" and defend their focus on this section of the literature. The term "argument-based literature" is not defined until the methods section on p. 7, lines 26-28, where they explain it as "an article using ethical concepts derived from current or traditional ethical theories". Merely "using" ethical concepts does not make an article argument-based - it needs to use them to make an argument in defence of a position or conclusion.

Answer from the authors: Meeting this request, we have added some further clarification in the methods section.

p. 7: [...] publication is considered to be in the argument-based literature, which is an article using ethical concepts derived from current or traditional ethical theories in order to argue for a position or conclusion […]

Additionally, the focus on argument-based literature (sometimes referred to as normative literature, e.g. on p. 5 line 29) requires justification.

Answer from the authors: The indistinct use between ‘argument-based literature’ and ‘normative literature’ seemed to be confusing. Following the reviewer’s suggestion, we have opted for the first term and use it consistently throughout the manuscript.

Presumably there is empirical bioethical literature that makes a relevant contribution to at least the first research question. Indeed, the authors mention that empirical studies have advocated for relational approaches to autonomy (p. 4, lines 27-29). Given the emphasis on understanding relational autonomy to improve end-of-life decision making in real-world situations, it seems that the empirical ethical literature might have something relevant to contribute.

Answer from the authors: We agree that empirical literature has much to contribute, indeed. Nevertheless, our interest was mainly focused on theoretical concepts and argumentations. Therefore, a choice in favour of argument-based literature was done. Of course, another
interesting research project may properly be to conduct a systematic review of the use of the concept of relational autonomy in the empirical literature.

Summary:

Overall this paper is of a high quality and makes an important contribution to the literature on autonomy in bioethics and decision-making in end-of-life care. This reviewer's recommendation is to accept the paper, but to request that the authors clarify how they understand "argument-based literature" and to defend the focus of their systematic review on the normative ethical literature.

Answer from the authors: thank you for the positive appreciation.