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)	Content and Outcomes of Narrative Medicine Programs: A Systematic Review of the Literature through 2019
AUTHORS	Remein, Christy; Childs, Ellen; Pasco, John; Trinquart, L; Flynn, David; Wingerter, Sarah; Bhasin, Robina; Demers, Lindsay; Benjamin, Emelia

VERSION 1 – REVIEW

REVIEWER	Antonietta Cappuccio
	Fondazione ISTUD
REVIEW RETURNED	07-Jun-2019
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GENERAL COMMENTS	I found interesting this manuscript and worth oh publication. On the other hand, all the narrative medicine definition are based on the American definition of narrative medicine, therefore the chosen criteria limited the identified publication to the ones that referred to Charon Approach.
DEVIEWED	Frie D. Marrous MD
REVIEWER	Eric R. Marcus, MD
DEVIEW DETUDNED	Columbia University USA
REVIEW RETURNED	11-Jun-2019
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GENERAL COMMENTS	They might want to make some comment about the quantitative study of a qualitative endeavor. It brings up the issue of how best to study qualitative social science variables and the amazing fact indirectly revealed that most of the studies done on this topic are quantitative.
REVIEWER	Jan W. Schoones
	Leiden University Medical Center, Walaeus Library
REVIEW RETURNED	29-Jul-2019
GENERAL COMMENTS	I am not a subject specialist - I am an information specialist. So my critique is limited to the literature search methods.
	The search strategy appears to be too limited, especially for the medical databases. If we look at PubMed, there seems to be no problem: the MeSH "Narrative Medicine" will be included when searching "narrative medicine"[all fields]. But, the MeSH "Narration" will not. Two problems. First of all, the MeSH "Narration", which includes itself the more specific MeSH "Narrative Medicine", appears to be a relevant subject heading - the definition for this MeSH is: "The act, process, or an instance of narrating, i.e., telling a story. In the context of MEDICINE or ETHICS, narration includes relating the particular and the personal in the life story of an individual." And when using this subject heading in a medical

database such as PubMed, the chances are that many references might potentially be relevant for the medical meaning of narration.

Second problem: the MeSH "Narrative Medicine" has been only recently been introduced in PubMed (in the year 2018; "Narration" was introduced in 2003), so one will miss references on the subject which do not use the phrase "narrative medicine" but do have the meaning of "narrative medicine". Adding the MeSH "Narration" to the query, the results will increase from 603 references to 8.195. An even larger increase will result when adding "narration" as a free text word as well (adding "narration"[tw] OR "narrations"[tw] OR "narrating"[tw] will increase the PubMed set to 8.818 references). So, there are in PubMed more than 8,000 references on Narration which have not been identified before, but which might include relevant items. It would be important to find a way to identify those possible missed relevant references by further specifying the search. For example, do the authors find the article The Use of Narrative as a Treatment Approach for Obesity: A Storied Educational Program Description (https://www.ncbi.nlm.nih.gov/pubmed/26999709) relevant? This article can be identifief by adding "program" as a second component of the query. The query might then be modified, for example, as: ("narrative medicine"[all fields] OR "reflective writing"[all fields] OR (("Narration"[Mesh] OR "narration"[tw] OR "narrating"[tw] OR "narrations"[tw]) AND ("program"[all fields] OR "programs"[all fields] OR "programme"[all fields] OR "programmes"[all fields]))).

Also, the original search is now 18 months old, so a reconsideration and the re-execution of the search might solve this issue as well.

REVIEWER	Rita Charon
	Columbia University, Medical Humanities and Ethics
REVIEW RETURNED	01-Oct-2019

GENERAL COMMENTS

This is a useful, high-altitude survey of a plethora of "programs" related in some way to the concepts and discipline of Narrative Medicine. There is heterogeneity in what is deemed a "program" from one short workshop to extensive longitudinal training. The strength of the manuscript is in its realization of the many outcomes that may be traceable to narrative medicine training. The weakness is the same. That is, the paper suggests something that might have been written about diabetes before diabetes was understood to be caused by high blood sugar: the method is successful in addressing polydipsia, polyuria, yeast infections, dry mouth, hunger, fatigue, blurred vision, etc. See? NM addresses burnout, lack of confidence, cultural competence, empathy, resilience etc, etc. What might the underlying foundational competencies be?

I suggest that the authors include more comprehensive descriptions of the well-described discipline of narrative medicine. The originators of the field have written a textbook and have published extensive discussions of the philosophical and literary-theory bases of the practice of narrative medicine. I see nowhere in this paper reference to the importance of phenomenology, intersubjectivity, literary reading theories, aesthetic theory of creative writing, or critical race theory, to mention a few of the fundamental conceptual frameworks of narrative medicine. Without

this grounding in theory, each "program" is being allowed to determine what the discipline is. Of course, the outcome is this plethora of outcomes that seem traceable to what is not, upon inspection, a unitary thing.

A proper literature review of this topic would have to include not only clinical literature but literary publications. Such journals as Narrative, PMLA, and Poetics Today have increasingly been publishing essays about narrative medicine. Perhaps the medical librarians could consult with librarians in Arts and Sciences to locate some of these resources too.

I believe this review is a valuable addition to the medical educational literature, and I applaud the authors for their careful survey and assessment of these publications. I believe the essay could be far more influential with a more comprehensive grounding in the intellectual/conceptual bases of the discipline itself. The "checklist" in Supplemental Appendix 6 would have to be revisited to suggest the clear proposition of a conceptual framework first before the "smaller" details of assessment and evaluation.

VERSION 1 – AUTHOR RESPONSE

Reviewer(s)' Comments to Author:

Reviewer: 1

Reviewer Name: Antonietta Cappuccio

Institution and Country: Fondazione ISTUD Please state any competing interests or state 'None

declared': None declared

Please leave your comments for the authors below.

I found interesting this manuscript and worth oh publication. On the other hand, all the narrative medicine definition are based on the American definition of narrative medicine, therefore the chosen criteria limited the identified publication to the ones that referred to Charon Approach.

Authors' response: Thank you for this insightful feedback. We agree that working definitions for narrative medicine (NM) which have evolved since the inception of the term (in 2001) frequently generate in the United States and therefore reflect a particularly American viewpoint on academic health sciences education. However, we have intentionally designed our systematic review to provide an inclusive overview of NM programming worldwide. We are pleased that the systematic review results includes programming based across the globe, including Nepal in 2009, the United Kingdom in 2010, Canada and Chile in 2012, France in 2013, Italy in 2014, Germany and Portugal in 2016, Iran in 2017, and Canada and China in 2018. (p. 22).

We have updated our Limitations section to note: "Since the conceptual framework of NM generated in the United States—although the framework is firmly rooted in an older tradition of medical and health humanities—to some extent the vocabulary and definitions associated with NM programming tends to reflect a particularly American viewpoint on academic health sciences education. We attempted to render our analysis of the records more globally inclusive by including programs that did not specifically employ NM language (some refer instead to "storytelling," "medical humanities," etc.) but still met our stated selection criteria." (p. 23).

Reviewer: 2

Reviewer Name: Eric R. Marcus, MD

Institution and Country: Columbia University USA Please state any competing interests or state 'None

declared': None

Please leave your comments for the authors below.

They might want to make some comment about the quantitative study of a qualitative endeavor. It brings up the issue of how best to study qualitative social science variables and the amazing fact indirectly revealed that most of the studies done on this topic are quantitative.

Authors' response: Thank you for providing this feedback. We agree that it is important to note the limitations involved with evaluating a qualitative endeavor in a field where quantitative methods frequently are employed for data interpretation. The Discussion section now reports that: "The qualitative studies highlighted a more nuanced breadth of outcomes regarding personal and professional benefits for participants in NM programs" (p.17). The Limitations section has been updated to note: "In a discursive way, our review also raises questions regarding the need for further study on best practices for performing quantitative studies of qualitative endeavors such as NM programming and other social science variables" (p. 24).

Reviewer: 3

Reviewer Name: Jan W. Schoones

Institution and Country: Walaeus Library, Leiden University Medical Center, Leiden, The Netherlands Please state any competing interests or state 'None declared': None declared

Please leave your comments for the authors below.

I am not a subject specialist - I am an information specialist. So my critique is limited to the literature search methods.

The search strategy appears to be too limited, especially for the medical databases. If we look at PubMed, there seems to be no problem: the MeSH "Narrative Medicine" will be included when searching "narrative medicine"[all fields]. But, the MeSH "Narration" will not. Two problems. First of all, the MeSH "Narration", which includes itself the more specific MeSH "Narrative Medicine", appears to be a relevant subject heading - the definition for this MeSH is: "The act, process, or an instance of narrating, i.e., telling a story. In the context of MEDICINE or ETHICS, narration includes relating the particular and the personal in the life story of an individual." And when using this subject heading in a medical database such as PubMed, the chances are that many references might potentially be relevant for the medical meaning of narration.

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Also, the original search is now 18 months old, so a reconsideration and the re-execution of the search might solve this issue as well.

Authors' response: Thank you for undertaking such a close reading of our manuscript and for your detailed and thoughtful comments.

The final search documented in our paper was certainly not the first – it was the result of a careful consideration of best practices, the scope of the research, and an analysis of previous iterations. As you note, other MeSH Terms could be added to our search strategy and they were present in the initial drafts. For example, "Narration" had been considered, yet we found that too many of the results were irrelevant given the goal of our project (possibly due to the absence of any didactic elements from the Scope Note's definition of the concept) even when paired with terms such as "Curriculum"; adding subheadings such as education often reduced the pool of results dramatically. We consulted both the Cochrane Handbook for Systematic Reviews of Interventions and the Joanna Briggs Institute Reviewer's Manual (specifically Chapter 2: "Systematic reviews of qualitative evidence") regarding sensitivity vs. precision and kept this paragraph from Cochrane in mind while revising the terms:

Searches for systematic reviews aim to be as extensive as possible in order to ensure that as many as possible of the necessary and relevant studies are included in the review. It is, however, necessary to strike a balance between striving for comprehensiveness and maintaining relevance when developing a search strategy. Increasing the comprehensiveness (or sensitivity) of a search will reduce its precision and will retrieve more non-relevant articles.

After consulting these resources we concluded that, while not perfect, "Narrative Medicine" [all fields] was our best choice going forward as it struck the best balance between sensitivity and precision. As you note this concept was not added as a MeSH Term until sometime after our search was conducted in 2017, which obviously precludes its use in our strategy. We have noted in our Methods section that this strategy: "we chose our search terms carefully in order to strike the optimal balance between sensitivity and precision." (p. 9).

We have examined newer citations, running subsequent searches following the original conducted in October 2017, with the most recent search being in October 2019. This 2019 search yielded an additional 305 citations, which we integrated into our earlier results. Of the 305 new citations, 16 qualified for inclusion in the systematic review. We have updated the results presented in our manuscript accordingly—please see Figure 1.

Reviewer: 4

Reviewer Name: Rita Charon

Institution and Country: Columbia University, USA Please state any competing interests or state

'None declared': None

Please leave your comments for the authors below.

This is a useful, high-altitude survey of a plethora of "programs" related in some way to the concepts and discipline of Narrative Medicine. There is heterogeneity in what is deemed a "program" from one short workshop to extensive longitudinal training. The strength of the manuscript is in its realization of the many outcomes that may be traceable to narrative medicine training. The weakness is the same. That is, the paper suggests something that might have been written about diabetes before diabetes was understood to be caused by high blood sugar: the method is successful in addressing polydipsia, polyuria, yeast infections, dry mouth, hunger, fatigue, blurred vision, etc. See? NM addresses

burnout, lack of confidence, cultural competence, empathy, resilience etc, etc. What might the underlying foundational competencies be?

Authors' response: Thank you for your insight in pointing out this limitation. We agree that this systematic review, which covers an array of diverse programs falling under the generally broad, and frequently ambiguously defined, umbrella of NM, is bound to discover and report a corresponding breadth of educational outcomes. We appreciate your pointing out the need for this explanation and have updated the manuscript conclusion accordingly: "Although a plethora of positive outcomes may stem from NM, its foundational competencies—as is implied by the very word humanities, from which NM draws its essential core—involve an ongoing exploration of what it means to understand reality and pursue human good, as this relates to our interactions with ourselves, others, and the world" (p. 25).

I suggest that the authors include more comprehensive descriptions of the well-described discipline of narrative medicine. The originators of the field have written a textbook and have published extensive discussions of the philosophical and literary-theory bases of the practice of narrative medicine. I see nowhere in this paper reference to the importance of phenomenology, intersubjectivity, literary reading theories, aesthetic theory of creative writing, or critical race theory, to mention a few of the fundamental conceptual frameworks of narrative medicine.

Without this grounding in theory, each "program" is being allowed to determine what the discipline is. Of course, the outcome is this plethora of outcomes that seem traceable to what is not, upon inspection, a unitary thing.

Authors' response: Thank you for your feedback. Although more detailed theoretical descriptions of NM as a discipline—and a robust delineation of the complex conceptual frameworks underling NM—is beyond the scope and purpose of this systematic review, we do appreciate your noting our oversight in failing to reference the textbook authored by originators of the NM field: Rita Charon, et al. The Principles and Practice of Narrative Medicine.

We now have included a reference to the textbook in the manuscript as a resource for readers who would like to pursue an in-depth exploration of NM's theoretical underpinnings: "The philosophical and theoretical underpinnings of NM as a framework for healthcare have been reviewed comprehensively by Rita Charon, et al. in The Principles and Practice of Narrative Medicine" (pp.5-6). We also have added the following to our Limitations: "we did not systematically analyze the philosophical and theoretical orientations of the specific NM program surveyed, and it is possible that doing so may have helped to inform our understanding of choices made in terms of program components and evaluations" (p. 23).

A proper literature review of this topic would have to include not only clinical literature but literary publications. Such journals as Narrative, PMLA, and Poetics Today have increasingly been publishing essays about narrative medicine. Perhaps the medical librarians could consult with librarians in Arts and Sciences to locate some of these resources too.

Authors' response: Thank you. While we appreciate that the scope of theoretical writings regarding NM is broad, we nevertheless chose to limit our selection criteria to databases that would include published literature focused primarily on the practical implementation of NM programming within academic health sciences professionals conducting clinical, educational, and research work. Our stated research question is: how effective is the implementation and evaluation of NM programs in academic medicine and health sciences? While humanities-based journals such as Narrative, PMLA, and Poetics Today do publish fascinating theory-based essays on the ideological significance, pedagogical framework, and interdisciplinary aesthetic of NM, articles documenting the implementation of specific NM programs at academic health sciences institutions typically are not

within the scope of these journals. Hence, we chose to limit our literature search to databases that primarily contain the types of studies that fit our selection criteria. More information regarding the design of our research strategy may be found in our previous response to Dr. Jan W. Schoones.

We are grateful for your insight, and have added the following to our Limitations section: "Because the scope of this review was focused on the implementation, components, and evaluation of NM programs, as opposed to the theory and philosophy underpinning NM as a conceptual framework for health, we did not extend our search to include humanities-based journals such as Narrative, PMLA, and Poetics Today" (p. 23).

I believe this review is a valuable addition to the medical educational literature, and I applaud the authors for their careful survey and assessment of these publications. I believe the essay could be far more influential with a more comprehensive grounding in the intellectual/conceptual bases of the discipline itself. The "checklist" in Supplemental Appendix 6 would have to be revisited to suggest the clear proposition of a conceptual framework first before the "smaller" details of assessment and evaluation.

Authors' response: Thank you for your thoughtful review of this manuscript; we appreciate your alerting us to the omission of such a section on NM theory in the checklist.

We have amended the checklist in Supplemental Appendix 6 to reflect the need for educators to consider the conceptual framework for NM in curriculum development and program implementation: "Program Design: Appreciate the theoretical underpinnings and conceptual framework of narrative medicine (NM) / \square Readings on NM Theory and Framework" (p. 20).

VERSION 2 – REVIEW

REVIEWER	Jan W. Schoones Walaeus Library, Leiden University Medical Center, The Netherlands
REVIEW RETURNED	27-Nov-2019

GENERAL COMMENTS	Thea authors have responded well to my review of their literature
	search.