PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	Strategies and impacts of patient and family engagement in collaborative mental health care: protocol for a systematic and realist review
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VERSION 1 - REVIEW

REVIEWER	Janet Treasure IOPPN, KCL, UK I have written self management books for individuals and families with an eating disorder
REVIEW RETURNED	28-Jun-2016

GENERAL COMMENTS	I was interested to read about this topic as I regard collaborative care to be of key importance and I was looking forward to the task of providing a review. However I am afraid that the language used in this protocol made my job as a reader very hard and took a great deal of time and additional research. Some of the technical terms used were not defined e.g. realist review. I therefore had to look up references and read other papers to get a basic understanding of the framing of this review. It was difficult for me to judge as a novice to this literature, whether their methods are indeed systematic and consistent with the convention for “realist reviews.” There was ambiguity and a lack of clarity about some terms which added to my difficulty e.g. sibling studies which is ambiguous and I thought initially were studies including siblings. Also “case summary “was used I think to describe the intervention again is ambiguous for a clinician. I would have thought that a reference to the TIDIER frame work which has been developed to classify interventions would be more appropriate for this. I found the description of how they would code and define the patient and family engagement and deliver which is a key part of the project unclear. The authors cite the frameworks of Coulter and Carman however when I went to these source papers I was not enlightened about the system/form of taxonomy to be used. It would perhaps be useful to have a diagram of a preliminary model of family/carer involvement so that it a clearer understanding of what theory needs developing is present. I found the description of the analysis to be quite vague. They mention that two authors will conduct a thematic analysis without

	really describing how that will occur. They then describe the "program theory refinement phase" in general terms. The authors do a good job describing how they will collect their data, involve stakeholders, etc. I do think that this will be a useful project and has the potential to make a helpful contribution to the literature but I did find the language and concepts confusing in part because I do not have a high level of expertise in qualitative methods however it would help if all the language was written in a clear, simple form.

REVIEWER	Melanie Gee Sheffield Hallam University, UK
REVIEW RETURNED	30-Jun-2016

GENERAL COMMENTS	General comments: This review protocol relates to two separate (but inter-related) reviews: a 'systematic', thematic review (part 1), and a realist review (part 2). Part 1 is a 'systematic' thematic review of studies investigating the effectiveness of different patient and family engagement strategies used in CMHC interventions for depression or anxiety disorders (i.e. did the strategies 'work'); part 2 is a realist review to develop and refine programme theories to explain why these different strategies (or a subset of these strategies) 'worked', for whom, and in what circumstances. The different review approaches and methods you have described for parts 1 and 2 are appropriate for answering your overall research question and your research objectives 1 and 2 respectively. However, the following issues should be addressed: Main comments: 1. Title and abstract. Given that part 1 does not relate to the realist review, but is a separate systematic thematic review (albeit one which does in part feed into the realist review), your title and abstract ('we are conducting a realist review') are misleading. 2. Outcomes. You acknowledge (p14) that in theory building for realist reviews it is helpful to work 'backwards' from key outcomes of interest. Identifying these key outcomes seems, to me, to be a crucial early stage of your realist review (part 2) and is an important part of focusing your review. When you state in your research question 'how do these strategies work', how do you/will you define what 'works'? Similarly in the objective 2, you refer to 'the particular outcomes triggered by these mechanisms' - which outcomes are you interested in, or how will you determine this? Will these be the same outcomes that you are using as inclusion criteria for part 1 (i.e. outcomes related to depression or anxiety symptomatology, medication use, functioning, quality of life, or satisfaction with care)? Or will you be led by your partners with lived experience to help select outcomes to focus on (as suggested on p14-15)? Or some combination of the two? This should be clarified. 3. Mechanisms. From your wording of objective 2 it appears that you may be inadvertently conflating the 'mechanism' of the CMO configuration with the engagement strategy per se. I agree that Dalkin et al (which you cite on p12) provides helpful advice here, in particular on p4: 'Differentiating between resource (the component introduced in a context) and reasoning therefore helps distinguish between relevant context and mechanism. Identifying the resource is

	contingent on the purpose of the study, and identifying the reasoning avoids the issue of conflating programme strategy (resource) with mechanism.' It would be helpful if you provided more explanation about what you, as a research team, understand by 'mechanism'. 4. Part 1 - eligibility criteria. It is not clear to me why your eligibility criteria (p7) do not additionally include studies relating to patient and family engagement strategies: these, presumably, are the only CMHC interventions that you will be interested in to address your objective 1. 5. Part 2 - search for initial theories. It would be helpful if you provided some more explanation (p9) about how you have used the pilot analysis of seminal articles on the topic of CMHC to identify the four patient and family engagement strategies you will use as a starting-point for theory building. 6. Part 2 - search for initial theories. I agree that content expert consultation and identifying topic-based theoretical and conceptual papers are good ways of finding relevant literature and insights to start theory-building (p9-10). However another potentially rich source of insights into why an intervention may or may not have worked would be the discussion sections of the relevant papers identified in part 1; similarly introduction sections often articulate the theoretical underpinnings of interventions. Realist review methodology requires that you engage with this body of literature in this way. 7. Part 2 - theorising. It is very common in realist reviews to produce more CMO configurations than can be practically tested and refined in later stages of the review. You should indicate (in broad terms) your planned strategy for prioritising theories for refinement should you find yourself in this situation. 8. Part 2 - search for evidence to refine theories. Cluster searching (p10) is an appropriate method for identifying sibling studies and associated evidence relating to previously identified relevant studies, in order to develop case studies to interrogate during theory refinement. However I am concerned that your starting point for the cluster searching is too limited: what is your rationale for restricting this to those studies identified in part 1, i.e. RCTs and clinical controlled trials? There will undoubtedly exist qualitative evaluations of patient and family engagement strategies which do not have a linked RCT or clinical controlled trial (e.g. Bentham, Wayne, et al. "Opportunities and challenges of implementing collaborative mental health care in post-Katrina New Orleans." Ethnicity & disease 21.3 0 1 (2011): S1). 9. Part 2 - quality appraisal. Conducting a formal study-level quality appraisal for those studies included in your realist review (part 2) has doubtful utility. It would be more meaningful to assess the rigour of the methods used to create the specific data from the study that you are interested in, and the relevance of that data to the theory that you are exploring (refer e.g. to the RAMESES training materials). 10. Synthesis. You should indicate how you will be presenting your synthesised results from parts 1 and 2. How will you be presenting your refined programme theories with supporting evidence? Will the two reviews be integrated in a final (narrative?) synthesis? A reference to the RAMESES reporting standards would also be

	helpful. Minor comments: 11. You refer to previous systematic reviews of CMHC (p5 line 24): these should be (re?-) referenced here. 12. The reference (p6 line 25) to the RAMESES guidance should have been to the RAMESES reporting standards. The correct reference is Wong, Geoff, et al. "RAMESES publication standards: realist syntheses." BMC medicine 11.1 (2013): 1. 13. Typographic error: p9 line 55, 'strategy' not 'strategies'.

VERSION 1 – AUTHOR RESPONSE

Reviewer 1	**Response**
I was interested to read about this topic as I regard collaborative care to be of key importance and I was looking forward to the task of providing a review. However I am afraid that the language used in this protocol made my job as a reader very hard and took a great deal of time and additional research. Some of the technical terms used were not defined e.g. realist review. I therefore had to look up references and read other papers to get a basic understanding of the framing of this review. It was difficult for me to judge as a novice to this literature, whether their methods are indeed systematic and consistent with the convention for "realist reviews."	We agree that there is a need to make the language used in the protocol more accessible to people that may be unfamiliar with realist reviews. We have provided a clearer definition of realist review and improved the language around its description on page 5 (2nd paragraph). The paragraph now states: "*Realist reviews offer an alternative approach to knowledge synthesis that is well adapted for complex models of care like CMHC. Realist reviews focus on providing explanations for how and why interventions may or may not work, for whom, and in what circumstances they work.*[29-31] *Such explanations are based on investigations into the mechanisms (i.e. causal forces) that underpin interventions and the contextual conditions impacting interventions' outcomes in different settings. The realist approach is considered theory-driven in that interventions are viewed as theories incarnate, made up of assumptions about how they are meant to work and what impacts they should have.*[31] *During the realist review process, these theories about how interventions generate change (also known as program theories) are made explicit and then compared against the existing evidence to find out whether they hold and are useful, or require refinement. Program theories thus map out the relationships between intervention contexts, components and mechanisms, and outcomes.*[31] *Realist reviews are interpretive in nature and typically draw from diverse sources of evidence, including quantitative, qualitative and mixed-methods research.*[29] *It is the appropriate method for helping us understand the complexity and potential value of patient and family engagement within CMHC*." We have also made attempts to better define terms used in the text (see next response).

There was ambiguity and a lack of clarity about some terms which added to my difficulty e.g. sibling studies which is ambiguous and I thought initially were studies including siblings.	We have gone through the text to improve the clarity of some terms that may be unclear to readers unfamiliar with realist reviews. The following changes have been made: On page 9 (paragraph 2): we specified that CMO stands for "context-mechanism-outcome" On page 10 (last paragraph): we clarified that sibling studies could refer to process evaluations or qualitative studies linked to the CMHC intervention trial) On page 12 (paragraph 2): we added a definition for 'middle range' theory as "theories that remain close to the data but also abstracted enough to apply to a broader range of situations"
Also "case summary "was used I think to describe the intervention again is ambiguous for a clinician.	In order to clarify what we mean by case summaries, we have provided an example on page 12 (3rd paragraph). We now state: *For example, relevant information from all studies, reports and other documents related to the IMPACT program for late life depression will be summarized to establish a detailed portrait of that particular case; the same exercise will be performed for all other CMHC interventions.*
I would have thought that a reference to the TIDIER frame work which has been developed to classify interventions would be more appropriate for this.	We thank the reviewer for this excellent idea, indeed we can use the TIDieR framework to better structure our case summaries. This point has been added on page 12 (2nd paragraph): *First, we will regroup all evidence related to each CMHC intervention and build 'case summaries' that provide rich descriptions of the interventions and the patient and family engagement strategies featured within them. These case summaries will be structured according to the TIDieR framwork[47] and provide a detailed account of interventions and their components, participants involved in delivering and receiving the interventions, activities related to patient and family engagement (materials, procedures, etc.), implementation contexts and processes, and reported outcomes.*
I found the description of how they would code and define the patient and family engagement and deliver which is a key part of the project unclear. The authors cite the frameworks of Coulter and Carman however when I went to these source papers I was not enlightened about the system/form of taxonomy to be used. It would perhaps be useful to have a diagram of a preliminary model of family/carer involvement so that it a clearer understanding of what theory needs developing is present.	We thank the reviewer for this comment and now include a preliminary engagement framework in the text (Figure 2). This framework (or typology) will be refined during the course of the review.

I found the description of the analysis to be quite vague. They mention that two authors will conduct a thematic analysis without really describing how that will occur. They then describe the "program theory refinement phase" in general terms.	We have added more description of our thematic analysis on page 9, paragraph 1: "*The thematic analysis will involve reading through the study articles, deductively and inductively establishing initial codes for specific engagement strategies (e.g. self-management supports, peer supports), and regrouping these codes into themes consistent with our conceptual frameworks (e.g. direct care strategies).*"
The authors do a good job describing how they will collect their data, involve stakeholders, etc. I do think that this will be a useful project and has the potential to make a helpful contribution to the literature but I did find the language and concepts confusing in part because I do not have a high level of expertise in qualitative methods however it would help if all the language was written in a clear, simple form.	We appreciate the reviewer's comments and have taken several steps to include the accessibility and clarity of the paper. We hope these steps are sufficient.
Reviewer 2	
1. Title and abstract. Given that part 1 does not relate to the realist review, but is a separate systematic thematic review (albeit one which does in part feed into the realist review), your title and abstract ('we are conducting a realist review') are misleading.	We agree that we have not emphasized the systematic review component of the review in the title and abstract. This has now been changed. The title now reads: "Strategies and impacts of patient and family engagement in collaborative mental health care: protocol for a systematic and realist review" In the abstract, we have modified the first sentence in the "Methods and analysis" section, which now reads: "We are conducting a review with both systematic and realist review components."
2. Outcomes. You acknowledge (p14) that in theory building for realist reviews it is helpful to work 'backwards' from key outcomes of interest. Identifying these key outcomes seems, to me, to be a crucial early stage of your realist review (part 2) and is an important part of focusing your review. When you state in your research question 'how do these strategies work', how do you/will you define what 'works'? Similarly in the objective 2, you refer to 'the particular outcomes triggered by these mechanisms' - which outcomes are you interested in, or how will you determine this? Will these be the same outcomes that you are using as inclusion criteria for part 1 (i.e. outcomes related to depression or anxiety symptomatology, medication use, functioning, quality of life, or satisfaction with care)? Or will you be led by your partners with lived experience to help select outcomes to focus on (as suggested on p14-15)? Or some combination of the two? This should be clarified.	We thank the reviewer for these questions. First, in review part 1, outcomes related to depression or anxiety symptomatology, medication use, functioning, etc. are part of our criteria for eligibility for intervention studies to be included in the systematic review update (this is stated clearly on page 7, last line of 3rd paragraph). These are often the types of outcomes most often reported in studies on CMHC. For review part 2, the outcomes that we will focus on will first be selected in partnership with our team members with lived experience of depression, as mentioned on page 15. The types of outcomes we will explore include clinical, service-level and implementation outcomes. We will then discuss these selections with the larger research team, making a decision together as a team. This has now been made explicit on page 15 (2nd paragraph): *"In a meeting with our partners with lived experience of depression, we will select outcomes to focus on from among the range of*

	outcomes we will be extracting data on (see section on 'data extraction and analysis' for review Part 1). These selections will be shared and discussed with the larger research team and a final decision on outcomes to focus on will be made."
3. Mechanisms. From your wording of objective 2 it appears that you may be inadvertently conflating the 'mechanism' of the CMO configuration with the engagement strategy per se. I agree that Dalkin et al (which you cite on p12) provides helpful advice here, in particular on p4: 'Differentiating between resource (the component introduced in a context) and reasoning therefore helps distinguish between relevant context and mechanism. Identifying the resource is contingent on the purpose of the study, and identifying the reasoning avoids the issue of conflating programme strategy (resource) with mechanism.' It would be helpful if you provided more explanation about what you, as a research team, understand by 'mechanism'.	We have made two changes to clarify our position on mechanisms. First, in our initial definition of the realist review on page 5 (paragraph 2), we mention the term 'mechanisms' and added *"(i.e. causal forces)"* to provide a basic definition for this term. Second, in our description of how we will conduct the realist synthesis on pages 12-13, we have provided examples for what we mean when we refer to mechanisms as intervention resources versus those related to reasoning or reactions (page 13, middle of the page). Specifically, we added: "Following advice from Dalkin, at this stage we will be sensitive to distinctions between mechanisms in the form of intervention resources (e.g. a self-management guide) versus those related to new reasoning or reactions (e.g. a patient's sense of self-efficacy)." We feel that these changes will help readers understand our position on mechanisms and show that we distinguish between visible intervention components and resources and the usually hidden mechanisms sought in CMO configurations.
4. Part 1 - eligibility criteria. It is not clear to me why your eligibility criteria (p7) do not additionally include studies relating to patient and family engagement strategies: these, presumably, are the only CMHC interventions that you will be interested in to address your objective 1.	The reviewer is correct in that the description of patient and family engagement strategies requires only the inclusion of CMHC interventions featuring these strategies. However, we did not add the presence of engagement strategies to the eligibility criteria for two reasons. Firstly, as CMHC intervention studies without engagement strategies can still in theory contribute contextual and possibly outcomes data relevant to our second objective (the realist synthesis), we did not want to exclude them during the systematic review update phase. Secondly, we are interested in knowing the extent to which different engagement strategies have been adopted across CMHC intervention studies. For example, out of all published CMHC interventions, how many adopted self-management strategies and how many adopted peer support strategies? This is a part of our intended descriptive analysis but was not initially mentioned in the text. We now mention our intent to quantify the extent of engagement strategies on page 9: "These analyses will help use to quantify the extent of different engagement strategies across CMHC studies and produce a rich description of these strategies as well as a typology based on

	our frameworks."
5. Part 2 - search for initial theories. It would be helpful if you provided some more explanation (p9) about how you have used the pilot analysis of seminal articles on the topic of CMHC to identify the four patient and family engagement strategies you will use as a starting-point for theory building.	We agree with the reviewer that few details were provided here. In fact, it is more accurate to say that our selection of four initial engagement strategies to focus on was based on both our pilot work as well as team discussions that we had while preparing the grant application for the review. We selected 15 studies included in a review of CMHC foundational studies written by Huffman and then performed a brief analysis in Excel to identify the engagement strategies featured in these studies. We found that out of the 15 studies in our sample, 11 featured either self-management strategies or patient involvement in treatment planning, or both. Only one study focused on peer or family involvement. However, our discussions as a team suggested that these latter strategies, though likely less commonly adopted, remained highly relevant for the realist review. Indeed, in the Cochrane systematic review by Archer, we know that there are several studies that make use of supports from peers and families. We have clarified these aspects on pages 9-10: "Informed by review team discussions and a pilot analysis of 15 seminal articles on CMHC identified in a review of foundational CMHC articles,[38] we have chosen to begin initial theory building with four patient and family engagement strategies appearing in CMHC interventions: shared decision-making and treatment planning, self-management supports, peer supports, and family supports. Patient involvement in treatment planning and self-management supports were featured in 11 of the 15 seminal articles we examined, suggesting that they may be commonly used strategies. Peer and family supports were much less commonly observed in the pilot analysis (adopted in only 1 study) but were identified by the review team as highly relevant for the realist review. Theory building will initially focus on these four strategies and continue in an iterative manner as other engagement strategies are identified in Part 1 of the review.
6. Part 2 - search for initial theories. I agree that content expert consultation and identifying topic-based theoretical and conceptual papers are good ways of finding relevant literature and insights to start theory-building (p9-10). However another potentially rich source of insights into why an intervention may or may not have worked would be the discussion sections of the relevant papers identified in part 1; similarly introduction sections often articulate the theoretical underpinnings of interventions. Realist review	We think this is an excellent idea and have added it to our methods on page 10: *"To locate and build initial theories related to patient and family engagement strategies we will: (1) consult with content experts in our review team and potentially outside the team if necessary, (2) perform electronic searches of the scientific and grey literature to retrieve theoretical and conceptual writings on each identified patient and family engagement strategies, (3) review the*

methodology requires that you engage with this body of literature in this way.	*introduction and discussion sections of articles on CMHC interventions retrieved in review Part 1, and (4) hold brainstorming sessions to create the CMO configurations and establish a consensus on these initial rough theories."*
7. Part 2 - theorising. It is very common in realist reviews to produce more CMO configurations than can be practically tested and refined in later stages of the review. You should indicate (in broad terms) your planned strategy for prioritising theories for refinement should you find yourself in this situation.	We thank the reviewer for this comment. Our CMO configurations will be prioritized in a full-team consensus building meeting that follows small group brainstorming sessions. This has now been added to the text on page 10 (last paragraph): *"Theory building will then advance through several small-group brainstorming sessions to construct CMO configurations for each engagement strategy and then these CMO configurations will be revisited and prioritized in a full-team consensus-building meeting supported by visualization software."*
8. Part 2 - search for evidence to refine theories. Cluster searching (p10) is an appropriate method for identifying sibling studies and associated evidence relating to previously identified relevant studies, in order to develop case studies to interrogate during theory refinement. However I am concerned that your starting point for the cluster searching is too limited: what is your rationale for restricting this to those studies identified in part 1, i.e. RCTs and clinical controlled trials? There will undoubtedly exist qualitative evaluations of patient and family engagement strategies which do not have a linked RCT or clinical controlled trial (e.g. Bentham, Wayne, et al. "Opportunities and challenges of implementing collaborative mental health care in post-Katrina New Orleans." Ethnicity & disease 21.3 0 1 (2011): S1).	The rationale for using the CMHC intervention studies as a starting point is that we will be creating case summaries for these interventions in order to have a stronger sense of how contexts may relate to mechanisms and outcomes. Each study linked to these RCTs or trials will contribute valuable information that helps us learn a little more about those specific intervention contexts as well as the particular engagement strategies used. However, we are clear in our paper that this is only a starting point and that we will not be limiting ourselves only to this literature. As mentioned on page 11 (paragraph 2), we state that we will also consider other studies of collaborative care where it can reasonably be inferred that the same engagement strategy mechanisms may be in operation. This is precisely where studies like the one cited by the reviewer will be identified and included in our realist synthesis.
9. Part 2 - quality appraisal. Conducting a formal study-level quality appraisal for those studies included in your realist review (part 2) has doubtful utility. It would be more meaningful to assess the rigour of the methods used to create the specific data from the study that you are interested in, and the relevance of that data to the theory that you are exploring (refer e.g. to the RAMESES training materials).	We agree with the reviewer and have thus modified our approach to remove emphasis on study-level quality assessment. The new sentences now read: "*Our appraisal of the quality of evidence used in the realist synthesis will be consistent with guidance by Pawson[28] and RAMESES standards.[32] Specifically, the rigour of each document or section of data will be appraised at the moment of their usage in the analysis. A subgroup within the review team with diverse methods expertise will establish minimal standards of quality for different study types (quantitative, qualitative and mixed-methods)*

	informed by the Mixed Methods Appraisal Tool (MMAT)[46] and meet together as needed to discuss the credibility and trustworthiness of other data sources relevant for the synthesis. The MMAT is a valid and reliable tool relevant for use in complex, mixed studies reviews as it contains criteria for appraising the methodological quality of studies with various study designs.[46] For example, if data relevant to our program theory were produced in a qualitative study, the MMAT suggests that the study is more trustworthy if the methods and analyses were described clearly and if the researchers performing the study explained how they may have influenced their results (i.e. reflexivity).”
10. Synthesis. You should indicate how you will be presenting your synthesised results from parts 1 and 2. How will you be presenting your refined programme theories with supporting evidence? Will the two reviews be integrated in a final (narrative?) synthesis? A reference to the RAMESES reporting standards would also be helpful.	While we think it is unlikely that we present the results from part 1 and 2 within a single final synthesis, we feel that at this early stage it is too early to tell precisely how we will be presenting results from each of the different parts of the review. We now make reference to the RAMESES reporting standards on page 6 (under Methods).
11. You refer to previous systematic reviews of CMHC (p5 line 24): these should be (re?-) referenced here.	We have added this references to the text (page 5, top paragraph 2).
12. The reference (p6 line 25) to the RAMESES guidance should have been to the RAMESES reporting standards. The correct reference is Wong, Geoff, et al. "RAMESES publication standards: realist syntheses." BMC medicine 11.1 (2013): 1.	We thank the reviewer, the correct reference has been added.
13. Typographic error: p9 line 55, 'strategy' not 'strategies'.	This error has been corrected (page 10, 2nd paragraph).

VERSION 2 – REVIEW

REVIEWER	Melanie Gee Sheffield Hallam University, United Kingdom
REVIEW RETURNED	21-Aug-2016

GENERAL COMMENTS	Thank you for your responses to my previous comments. I confirm that all the issues raised have been satisfactorily addressed in your responses and through amendments to the manuscript, and there are no outstanding matters requiring further attention. I wish you all the best with the study.